Towards the Prevention of Behavioural and Emotional Difficulties in People with Intellectual Disabilities.

A position paper on behalf of the Special Interest Research Group on Challenging Behaviour and Mental Health of the International Association for the Scientific Study of Intellectual Disabilities.

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Introduction

Both challenging behaviour (such as aggression, destructiveness and self-injury) and mental ill-health (for example, anxiety, depression, mania, and psychosis) are commonplace amongst people with intellectual disability. For example, research from the UK (Emerson & Hatton, 2007; Emerson & Einfield, in press), Australia (Emerson, Einfield, & Stancliffe, in press), Finland (Linna et al., 1999) and the Netherlands (Dekker et al., 2002) has suggested that between 20-30% of children with intellectual disability experience behavioural difficulties as compared to 4-10% of children without cognitive impairments. As such, children with intellectual disability account for approximately 15% of all children with a diagnosable mental health problem (Emerson & Hatton, 2007). Recent UK research reported a combined prevalence rate in an adult population of between 16-41% depending on the assessment techniques employed (Cooper et al., 2007). In the latter study, challenging behaviours accounted for between 33-50% of identified difficulties.

Applying such figures on a global basis is problematic. Prevalence studies on intellectual disability are typically conducted in high-income countries, and it cannot therefore be assumed that identified rates would also apply in lower-income nations. For example, while the overall prevalence of intellectual disability has been estimated to lie between 1-3%, the incidence of intellectual disability is likely to be substantially higher in low-income countries as a result of the increased exposure to a wide range of
environmental risks (Durkin, 2002; Emerson et al., 2007; Institute of Medicine, 2001; World Health Organization (WHO), 2002). Similarly, the identification of abnormal patterns of behavioural and emotional responding will vary as a function of both cultural differences in interpretation and service availability (Raghavan & O’Hara, 2005) and exposure to a range of environmental risk factors such as extreme poverty, disasters and armed conflict (Lancet Global Mental Health Group, 2007). However, assuming an average global prevalence rate for intellectual disability of 2%, and a prevalence rate for emotional and behavioural disorders of 20-30%, then between 27-40 million of the world’s citizens would be predicted to have an intellectual disability plus additional needs in terms of challenging behaviour or mental-ill health.

While the relationship between these two phenomena is complex and yet to be subject to definitive research (Allen, 2008; Emerson, Moss & Kiernan, 1999; Hemmings, 2007), the consequences of challenging behaviour and mental-ill health, either alone or in combination, can be particularly dramatic in this population. For persons with intellectual disability themselves, these can include social exclusion, institutionalisation, deprivation, physical harm, abuse, misdiagnosis, exposure to ineffective or aversive interventions, and failure to access evidence-based interventions (Baker & Allen, 2001; Emerson, 2001; Guess et al, 1987; Lowe et al., 2005; Rusch et al, 1986; White et al., 1995). Their carers face elevated risks of physical and mental-ill health, physical injury, increased financial burdens, and reduced quality of life (Allen et al, 2006; Qureshi, 1994). For those charged with commissioning and providing services, the major impacts include potential exposure to negative publicity resulting from scandals associated with poor care provision and having to fund escalating care costs (Department of Health, 2007). Given that challenging behaviours and mental ill-health may first appear in childhood (Einfield et al, 2007; Murphy et al., 1999) and, in the absence of appropriate intervention, often seem to be enduring (Einfield et al., 2006; Kiernan & Alborz, 1996), significant costs may be payable over protracted periods of time for some individuals. In the early
1990s, the National Institutes for Health (1991) estimated that 200,000 individuals with developmental disabilities in the United States displayed significant degree of destructive behaviour at an annual cost to care services exceeding $3 billion. Annual individual service costs of between 100-450,000 pounds sterling have recently been identified in the UK (Emerson & Robertson, 2008; Lowe et al, 2007a).

Although direct care costs in low-income countries will not match these levels due to the low availability and coverage of services, low treatment costs for behavioural and psychiatric problems may serve to inflate indirect costs by increasing the duration of untreated conditions and the resulting disability arising there from (WHO, 2004).

Challenging behaviour and mental-ill health in people with intellectual disabilities are therefore high-impact conditions that can have significant direct and indirect clinical, social, and fiscal implications. Conditions that have similar impacts within the general population (e.g. coronary heart disease, smoking-related illnesses) are typically subject to high-profile public health interventions whose focus is prevention. In contrast, behavioural and emotional difficulties in people with intellectual disabilities are often only addressed when they have become fully established in a person’s behavioural repertoire, present for many years, and therefore likely to be more resistant to effective intervention.

The prevention of any disease or condition requires the implementation of a potentially complex range of programmes and actions that may involve government agencies, professional organisations, public health professionals, individual clinicians, service providers, carers and potentially at risk individuals themselves. Three levels of prevention have traditionally been considered (WHO, 2004):

- **Primary prevention** involves impacting on the incidence of a condition by inhibiting its onset. Primary prevention strategies can be further sub-divided into universal prevention (interventions that are targeted at the general public or whole population groups that
have not been identified on the basis of increased risk), *selective prevention* (interventions that target individuals or population subgroups where the risk of developing a condition is significantly higher as a consequence of the presence of biological, psychological or social risk factors), and *indicated prevention* (intervention that target high-risk individuals identified as having minimal but detectable signs of a disorder but who do not reach diagnostic criteria for caseness at that point in time).

- **Secondary prevention** involves screening in order to detect a condition before it becomes fully symptomatic and the use of early intervention to reduce the probability of the full-blown condition developing. The theoretical principle here is that intervention takes place when it is most likely to be successfully treated; with early intervention, it may be possible to eradicate a condition, slow its progression, prevent or minimise complications and limit any resulting disability.

- **Tertiary prevention** focuses on individuals who already have an identified condition and strives to prevent further disability and restore pre-morbid function as far as is possible. Tertiary prevention programmes aim to improve the quality of life of those affected by again limiting complications and disabilities, reducing the severity and progression of a condition, and providing rehabilitation to restore functionality and self-sufficiency. Unlike primary and secondary prevention, the effects of which are essentially indirect, tertiary prevention involves the direct treatment of a condition.

Though this system has sometimes been criticised on the basis of a lack of distinction between these levels, and particularly between primary and secondary, this three-tier model remains in common usage.

This paper outlines a public health approach to behavioural and emotional difficulties in people with intellectual disabilities and suggests
strategic intervention opportunities at each of the three levels described above. This stance is congruent with the WHO’s (2004, 2007, 2008) perspective that both intellectual disability and mental health are public health issues.

While empirical evidence to support such a model is lacking at the present time, there is reasonably strong circumstantial evidence to suggest that preventative strategies may be effective in this population. Our aim in this paper is therefore to review the evidence that does exist and to consider the nature of the strategies that might be expected to be effective in reducing the incidence of behavioural and mental health problems amongst people with intellectual disability.

**Risk and Protective Factors**

Risk and protective factors are important concepts in developing preventative interventions. Risk factors are associated with an increased probability of onset, greater severity and longer duration of health concerns, while protective factors modify, alter or ameliorate an individual’s response to environmental hazards that predispose to ill health (WHO, 2004). Interest in the latter grew out of the acknowledgement that many individuals exposed to stress and adversity do not go on to develop difficulties later in life; this in turn prompted research into factors that enhance psychological resilience.

Rutter (1990) described four types protective factors: those that reduce exposure to risk; those that reduce negative chain-reactions following negative events or experiences; those that promote self-esteem and self-efficacy through achievement; and those that promote positive relationships and new opportunities that help increase personal resources and provide new life opportunities. Both risk and preventative factors can be generic across conditions or specific to a single condition, and, while underpinning mechanisms may vary, there is some evidence to suggest that resilience is a cross-cultural phenomenon (Gunnestad, 2006).
Social, environmental and economic risk factors for mental ill-health in the general population include: isolation and alienation, lack of education, peer rejection, poor social circumstances, poverty, social disadvantage, family contact and disorganisation, parental mental health issues, exposure to stressful life events and unemployment. Universal protective factors include: empowerment, positive interpersonal interactions, social participation, and access to good social support. Individual risk factors include academic failure, chronic health problems, and lack of social skills; in contrast, protective factors include adaptability, autonomy, good stress management skills, early cognitive stimulation, and good life skills (WHO, 2004).

People with intellectual disability will, in general, experience high levels of exposure to these known risk factors. For example, Emerson & Hatton (2007) showed that cumulative risk of exposure to a variety of indicators of social disadvantage (lone parent family, income poverty, exposure to two or more negative life events, poor family functioning, primary carer with no educational qualifications, potential maternal mental health issues, and poor maternal self-rating of health) were associated with increased prevalence of emotional disorders, conduct disorders and hyperactivity amongst children. While this was true for those with and without an intellectual disability, the former were at significantly greater risk of exposure to all the variables studied. People with intellectual disabilities are also at significant risk of experiencing social isolation (McVilly et al, 2006: Stancliffe et al., 2007), being unemployed (Martorelli et al., 2008), being supported in settings where there are low levels of activity and stimulation (Mansell et al, 2003) and, while they are at increased risk of experiencing a wide variety of general health problems, the treatment that they receive for these problems often falls below optimal levels (Scheepers et al., 2005). The latter is particularly important given the reciprocal influences that physical and mental health may exert upon each other; cardio vascular disease, for example, can result in depression and vice versa (WHO, 2004). Furthermore, the misdiagnosis of a medical illness as a behavioral disorder is a significant problem that can
impede the diagnosis of a potentially life-threatening condition and can expose individuals to unnecessary pain and suffering (Mikkelsen, 2007). Mikkelsen & McKenna (1999), for example, estimated that 5%-10% of individuals with intellectual disability who present with a significant negative change in their behavioral status may have an underlying medical illness contributing to the exacerbation in behaviour. Kennedy et al. (2007) also found that children with severe developmental disabilities and challenging behaviour had significantly higher rates of special healthcare needs compared to matched-comparison controls.

Life events are well-established precipitants of psychological distress in the non-learning disabled population. Murphy et al (2007) similarly found that behavioural difficulties in a group of people with intellectual disabilities increased following experience of abuse, while Hulbert-Williams & Hastings’ (2008) review of the literature on the impact of life events also reported clear associations with psychological problems.

People with intellectual difficulties will in general also fair poorly in relation to the presence of protective factors that promote resilience. Often they will be disempowered, experience low levels of social participation and receive poor social support. By virtue of their limitations in cognitive and adaptive functioning, many will also lack the personal protective factors that help insulate against the effects of these identified risk factors. Viewing these considerations in combination, it is perhaps small wonder that emotional and behavioural disorders are more prevalent amongst this population.

**Primary Prevention**

Seccombe (2002) differentiates between preventative initiatives designed to ‘change the odds’ and those designed to ‘beat the odds’ of a condition developing. The former are likely to involve policy level changes designed to avoid exposure to known risk factors in the first instance,
whereas the latter more typically involve group or individual level interventions designed to foster the development of protective capacity and resilience. In terms of the model described above, while primary prevention is essentially concerned with both ‘changing the odds’ by reducing exposure to risk) and ‘beating the odds’ (by building generalised resilience, secondary and tertiary prevention focus solely on ‘beating the odds’. Universal macro-strategies known to improve mental health and reduce the risks for mental ill health in the non-intellectually disabled population include: improving nutrition, housing, access to education, reducing economic insecurity, strengthening community networks and reducing harm from addictive substances (Anderson et al., 2005; Costello et al., 2003; Patel & Jané-Lopez, 2005).

There is a growing evidence base regarding the efficacy (and to an extent, the effectiveness) of primary preventative interventions in the general area of child and adolescent mental health (Boisjoli, Vitaro, Lacourse, Barker, & Tremblay, 2007; Dishion et al., 2008; Offord & Bennett, 2002; Tremblay, 2006; Webster-Stratton et al., 2008; Webster-Stratton & Taylor, 2001). Congruent with the information above, most of these strategies seek to improve the developmental environment in which children grow up by either reducing the risk of exposure to adverse environmental conditions (i.e., by changing the odds) or by promoting child, family or community resilience in the face of adversity (i.e., by beating the odds) (Goldstein & Brooks, 2006; Luthar, 2006, 2003; Luthar & Brown, 2007).

Given that reported associations between environmental adversity and mental health are similar for children with and without intellectual disability, it appears plausible to suggest that the efficacy of primary preventative interventions in the general area of child and adolescent mental health should generalise to children with intellectual disability. Given the association between social deprivation and challenging behaviours amongst children with intellectual disability described above, universal initiatives such as the Child Poverty Bill in the UK, which aims to eradicate child poverty by 2020, if
successful, should also have a positive impact on the incidence of such behaviours.

A number of selective interventions should also have a positive impact. The movement of people from large-scale institutional settings into smaller scale community living arrangements has been shown to be generally associated with improved material standards of living, greater satisfaction, more opportunity to use skills, increased time in engagement in constructive activities, increased contact with other people and a greater ability to exercise choice (although people also remain relatively poor, develop relatively few new skills post-move, and still spend relatively long periods unoccupied).

While the evidence is that such transitions can in themselves have positive impacts on stereotypic behaviours (such as rocking, finger flapping etc.), they do not appear to impact greatly on more serious challenging behaviours (Emerson & Hatton, 1994). This is perhaps not surprising, as more established behaviours are likely to require secondary or tertiary level intervention strategies. It remains plausible, however, that individuals without previous institutional experiences may be less likely to develop behavioural and emotional difficulties if all of their experiences are of high-quality community living environments. The development of work opportunities via supported employment programmes, which have a number of demonstrable benefits including increasing self-esteem, satisfaction and engagement in meaningful activity (Beyer & Robinson, 2009), are also likely to have positive impacts on psychological well-being.

Finally, there are a number of demonstrations that the general health status of people with intellectual disabilities can be improved via the introduction of routine health screening checks, which again may have beneficial indirect impacts on behavioural and emotional functioning (Webb & Rogers, 1999; Baxter et al, 2006; Cobb et al, 2008). This is extremely important as, in addition to the established association between physical and mental wellbeing in general (WHO, 2004), there numerous ways in which physical health problems can manifest as or set the scene for the occurrence
of challenging behaviours and mental health problems in people with intellectual disability (and most notably amongst those with higher levels of disability) (Kennedy & Becker, 2006; Baker, 2009; Gouras, 1986; Lennox, 2007).

There are also more specific ways in which such primary prevention strategies could impact on emotional and behavioural problems in this group. There are, for example, a number of known genetic and environmental influences that cause intellectual disability per se, but which also impact on later emotional and behavioural presentation. For example, the ingestion of a range of legal and illicit drugs during pregnancy can have teratogenic effects on the foetal nervous system. Anticonvulsants, particularly sodium valproate (Motamedi & Meador, 2006) and psychotropic drugs such as antidepressants and anxiolytics taken during pregnancy (Kallen, 2004; Bercovici 2004) are associated with subsequent cognitive behavioural and neurodevelopmental problems. Maternal alcohol consumption, smoking and the use of substances such as marijuana, cocaine and heroin, have obvious or more subtle effects on subsequence cognitive function, behaviour and development of the child. Consumption of alcohol during pregnancy produces a range of subsequent developmental behavioural mental health and attachment problems referred to as the foetal alcohol spectrum disorder (FASD) (Nulman et al., 2007). The prevalence of FASD is much more common than that of Down’s syndrome and is estimated to affect at least 1% of children although it is more frequent in indigenous children, for example, in Australia with a rate of 2-5% or in other populations where high consumption of alcohol is common (Abel 1998: Harris & Bucens, 2003; May et al, 2005). International consensus is that abstinence from alcohol during pregnancy is necessary to prevent FASD (Nulman et al 2007; NH& MRC, 2009). Strategies which lead to lower rates of exposure to such toxins should therefore have a specific impact on the prevalence of emotional difficulties as well as on intellectual disability per se.
While not as well established as the research concerning the teratogenic effect of neurotoxic exposures, there is a growing body of evidence that exposure to a variety of natural and man-made chemicals may play a part in the development of certain psychiatric symptoms or disorders. Brown (2002, 2007) reports psychiatric sequelae of exposures to a number of toxins, including insecticides; metals such as lead, arsenic, mercury, and tin; solvents such as gasoline, benzene, toluene, glue, and paint; and polybrominated biphenyls and polychlorinated biphenyls. A precautionary approach to chemical regulation will help reduce the incidence of these exposures and mitigate the injury caused (Gilbert, 2008).

Preventative interventions designed to reduce the impact of stressors and enhance resilience include promoting positive starts to life (via home-based programmes and pre-school educational and parenting interventions), reducing child abuse and neglect, providing effective support for parental mental health issues, responding positively to family disruption and by building resilience and reducing risk behaviours in schools.

General population home intervention programmes that focus on improving social support, parenting skills and parent-child interactions during pregnancy and early infancy have demonstrated gains that include improved mental health of mothers and children, less use of health services and long-term reductions in challenging behaviours (Brown & Sturgeon, 2005). Importantly, there is some evidence that family based interventions of this type can be effective in low-income and middle-income countries (Patel et al, 2007). The value of such early intervention programmes for children with intellectual disability is well recognised (Guralnick, 2005). Given that absent or limited social contact outside of the family in children with intellectual disability has been found to predict mental-ill health in young adult life (Tonge & Einfield, 2003), activities that encourage the development of social relationships may also help insulate against such the development of emotional problems.
Similar preventative strategies have been shown to be effective in generic school settings. Crone & Horner (2003), for example, describe how positive behavioural support\(^1\) can be utilised on a whole-school basis to build the resilience referred to above. In their model, they estimate that this would involve approximately 80-90% of students being exposed to universal prevention via, for example, their being taught social skills and being governed by common reinforcement systems; the residual 10-20% would require more focussed secondary and tertiary prevention interventions. There is some emerging evidence that such models can be highly effective with non-intellectually disabled children (Horner et al, 2009; Bradshaw et al 2009). In combination with effective home-based intervention, the systemic application of such approaches within both integrated and specialist educational facilities for children with intellectual disability could have a major preventative impact on the incidence of emotional and behavioural difficulties in this population also.

Carr et al. (1998) distinguished between *molecular* behavioural interventions (which concentrate on understanding and altering the immediate antecedents and reinforcers for behaviour) and *molar* behavioural interventions (which focus on much broader aspects of the environment, such as living and family arrangements, but which also impact on behaviour in a more global sense). Whereas the interventions described above are essentially molar in nature, applied behaviour analysis has more traditionally focused on molecular level analysis and intervention. Based on this literature, micro level primary prevention strategies would involve identifying more immediate environmental events that increase or reduce the probability of subsequent challenging behaviour; a potential primary prevention strategy would therefore be to reduce the frequency of the former and increase the frequency of the latter. For example, it is known that demands made by

\(^1\) An approach to behavioural intervention that rejects the use of aversive interventions that are dependent on punishment contingencies in favour of antecedent and reinforcement based strategies (Guess et al., 1987; LaVigna, Willis & Donnellan, 1989; Meyer & Evans, 1989; Luiselli, 2006). Although developed initially within the intellectual disabilities field, its use has now extended to other populations.
carers are one of the most common factors provoking incidents of challenging behaviour (e.g., Iwata et al., 1994). As part of a preventative approach, carers could therefore be taught about the relationship between demands and challenging behaviour and why it exists. This might lead to questioning the necessity of some demands, but also to teaching them how to use some of the very wide range of approaches that have been developed through work with individuals to prevent demand-related challenging behaviour. These include, for example, routinely giving people more choice about what to do and when to do it (Dyer et al., 1990), embedding demands in a positive social context (Carr et al., 1994), preceding hard with easy demands (Mace et al., 1988), giving notice that a demand is on the way (Tustin, 1995) and so on. Outside of the context of highly structured behavioural programming, such approaches are little used (or even known), yet they are relatively easy to implement and likely (at the population level) to be effective at preventing a proportion of new incidents of challenging behaviour.

This approach relies on an ability to identify common factors underlying behavioural episodes. As well as demands, it is known, for example, that incidents are more common in circumstances of social deprivation, when food or other preferred items are restricted, and when the environment is barren and unstimulating (McGill, 1999). Just as with the above analysis of demands, these factors can be unpacked and arrangements made to ensure that environments are less socially deprived, preferred items and activities are more available and environments are arranged to be as stimulating as possible, potentially via the routine application of proven interventions such as Active Support (Stancliffe et al., 2008). Although the research evidence is somewhat equivocal, there are a number of studies that indicate that the introduction of this approach can indeed have beneficial impacts on rates of challenging behaviour (Koritsas et al, 2008; Toogood et al, 2009). Conversely, for persons on the autistic continuum, highly structured environments that offer lower levels of arousal will be indicated (Clements, 2005).
Many other micro-environmental factors are likely to be implicated in the occurrence of challenging behaviour. Some of these (e.g. overcrowding, noise, change etc.) are already known or suspected, while others no doubt remain to be discovered. This suggests then a new approach to epidemiology in which the focus is not just on the characteristics of the individuals who are most frequently involved in incidents of challenging behaviour, but also on the ecological factors associated with those very incidents. Attempts to construct such a knowledge base have begun (McAtee et al., 2004; McGill et al., 2003) but require significantly more research.

Secondary Prevention

Intervention at this level may hold great promise for the prevention and early remediation of behavioural and emotional difficulties in people with intellectual disabilities (Schroeder et al., 1986). As stated above, number of studies suggest challenging behaviours typically emerge at a relatively young age in this population (e.g., Berkson et al., 2001; Kurtz et al., 2003; Richman & Lindauer, 2005; Schneider et al., 1996). For example, a retrospective study examining the origin of self-injurious behaviour (SIB) in people with intellectual disability found that 68% began displaying such behaviours before the age of 5 years (Schneider et al., 1996). Once established in the child’s repertoire and in the absence of effective intervention such behaviours tend to be characterised by their chronicity (Murphy et al., 2005); indeed prevalence studies suggest an increase in the frequency and severity of challenging behaviour in people with intellectual disability between the ages of 5-25 years (see Oliver, Murphy, & Corbett, 1987).

Current understanding of the genesis of challenging behaviour suggests that early behaviour-environment relationships are critical to its development. Existing conceptual models of the development of SIB suggest that such behaviours begin as relatively ‘uncommitted’ responses. These early behaviours come into contact with socially and non-socially mediated
contingencies of reinforcement and become operant responses (Guess & Carr, 1991; Kennedy, 2002; Richman, 2008). Over time, caregivers become less responsive to these precursor behaviours, leading to an increase in the frequency or severity of the child’s behaviour, which then re-evokes the reinforcing response from caregivers. This process effectively shapes not only the development of the child’s challenging behaviour, but also caregiver responses to it (Oliver, 1993; 1995). Some empirical data now exists to support this mutual reinforcement paradigm in groups considered ‘at risk’ of developing SIB (Hall et al., 2001; Oliver et al., 2005; Richman & Lindauer, 2005). Although this work has focused exclusively on self-harm, it seems likely that the differential reinforcement of precursor behaviour is important for the development of all topographies of challenging behaviour.

Studies of the epidemiology of challenging behaviour have suggested a number of personal characteristics that act as risk markers for its development (McClintock et al., 2003). The following are all positively correlated with the presence of challenging behaviour: being male (e.g., Maurice & Trudel, 1982), having a severe or profound intellectual disability (e.g., Chadwick et al., 2008; Jacobson, 1982), deficits in adaptive behaviour (e.g., Hartley et al. 2008), sensory and physical impairment (e.g., Kiernan & Kiernan, 1994), having a diagnosis of autism (e.g., Baghdadli et al., 2008; McClintock et al., 2003), delays in either expressive or receptive communication (e.g., Schroeder et al., 1978), and the presence of certain genetic conditions, such as Lesch-Nyhan (Anderson & Ernst, 1994) and Cri-Du-Chat syndrome (Collins & Cornish, 2002). Furthermore, some phenotypes are known to be associated both with specific forms of challenging behaviour and particular forms of functional psychiatric disorder; individuals with Prader-Willi syndrome, for example, are likely to engage in skin picking at specific body sites (Thompson & Caruso, 2002) and to experience psychosis (Boer et al, 2002 ) and affective disorders (Soni et al, 2007). Knowledge of specific psychopathology that is associated with a behavioural phenotype clearly has implications for early intervention and
treatment. For example, modified cognitive behavioural therapy and/or anxiolytics may have great utility in young people with Williams syndrome (a deletion on the elastin gene section of chromosome 7) who typically develop severe anxiety (O'Brien & Yule, 1995).

At present little is known about the process by which the risk markers described above influence susceptibility to behavioural and emotional difficulties and, more specifically, how they influence the early behaviour-environment relations that appear to be necessary for the development of challenging behaviour\(^2\). It seems likely that there are multiple developmental pathways by which challenging behaviours become established in the person's behavioural repertoire (Richman, 2008). The presence of some of the above risk factors may serve to potentiate the developmental process described above. It has, for example, been suggested that gene-environment interactions may play a critical role in the development of challenging behaviour for some individuals with intellectual disabilities (Kennedy et al., 2001; Langthorne & McGill, 2008). Recent studies have suggested that individual gene variations found among the general population placing people at-risk for challenging behaviour (Caspi et al., 2002) may also play a role in the development of aggressive and/or self-injurious behaviors in people with intellectual disabilities. For example, functional polymorphisms in the monoamine oxidase A promoter gene and/or serotonin transporter gene have been associated with challenging behaviors (Cohen et al., 2003; Hessl et al., 2008; May et al., in press). These findings regarding genes involved in brain circuit dysfunction may play a role in the development of challenging behavior, particularly when additional environmental stressors are present (Taylor & Kim-Cohen, 2007).

Screening tools are required that enable the identification of those most ‘at risk’ of developing challenging behaviour at a sufficiently young age. Such tools need to take account of the multiple risk markers described above

\(^2\) See Kraemer et al. (2001) for a detailed account of the ways in which risk factors may impact on behavioural and mental health outcomes
and need to be sufficiently accurate to justify targeted interventions, as the identification of only those most at risk may miss the majority of individuals who go on to develop behavioural difficulties. Although there is no agreed upon standard for positive predictive value in screening, Bennett et al (1999) have suggested that screening procedures should have a sensitivity of at least 50%. In some cases, screening would be relatively clear. For example, all children who receive a diagnosis of a genetic condition correlated with particular challenging behaviours could be referred for early assessment and intervention following initial diagnosis (Langthorne & McGill, 2008). In other cases, such indicators may not be so apparent; and more subjective screening tools may also be required to supplement this process. For example, Murphy et al (1999) found that the most effective predictor of the subsequent development of SIB was the level of teacher concern for the child. Einfield & Tonge (2003) found that high levels of emotional and behavioural difficulties in young children with intellectual disability was a very significant independent predictor of ongoing problems into adulthood.

Functional behavioral assessment is considered best practice for the assessment of challenging behaviour and a large body of literature exists to show that challenging behaviours displayed by people with intellectual disabilities are frequently maintained by both social and non-social consequences (Hanley et al., 2003). Interventions that are derived from this understanding, such as functional communication training (FCT), are more likely to lead to clinically significant and socially valid change than those based on alternative rationales (Pelios et al., 1999).

Such methods can be adapted for use with young children with intellectual disabilities who are at risk of developing serious challenging behaviour and may require briefer, less intense implementation than would be required with more fully-established behaviours (Crone & Horner, 2003); Kahng & Iwata (1999), for example, have shown that briefer forms of experimental functional analysis can be as reliable as longer forms in about two thirds of cases.
A small number of studies have demonstrated the effectiveness of adopting a function-based approach to early intervention with this population (Kurtz et al., 2003; Reeve & Carr, 2000; Wacker et al., 2005; Wacker et al., 1998). Wacker et al (1998) trained a group of 28 parents to conduct a functional analysis of challenging behaviours displayed by their young child (aged 1-6 years). A motivating consequence was successfully identified for 89% of the children, with some 86% of children displaying challenging behaviour that served a clear social function. Parents were then trained to implement a FCT intervention for their child’s behaviour and the effectiveness of this intervention was monitored at 3 monthly intervals for up to 12 months. The average pre-post decrease in challenging behaviour for all participants was 87% (range 45%-100%); there were also collateral increases in positive child social behaviours. Parents rated this model of early intervention as highly acceptable and as causing minimal disruption to family life. Wacker et al. (2005) found similar results with a second group of 25 young children (aged 1-7 years) who displayed challenging behaviour. Reductions in challenging behaviour were also shown to generalize across untrained settings, tasks and people. Studies by other research groups have reported equally promising findings (Kurtz et al., 2003; Reeve & Carr, 2000). The latter study compared FCT against expressive language training (ELT) in 8 young children (aged 2-5 years) who displayed minor challenging behaviours maintained by attention. The four children who received a FCT treatment showed reductions in challenging behaviour and an increase in communicative behaviours, in contrast the four children who received ELT showed increases in challenging behaviours until receiving the FCT treatment.

The benefits of active caregiver involvement have been demonstrated in early intervention programmes for children with autism (McEachin et al., 1993). A similar level of caregiver involvement may be required if the benefits of early intervention for challenging behaviour are to generalize across time and settings (Dunlap & Fox, 1996). Adopting a consultation model of service
delivery, similar to that used by Wacker et al., that equips caregivers with expertise in their child’s challenging behaviour may help to ensure that the advantages of early intervention maintain as intensive early support is withdrawn and the child enters new environments.

Of particular relevance is evidence that effective early behavioural intervention strategies can be delivered on the scale required to make an impact at a whole population level. Matthews et al. (2009), for example, described the widespread implementation of a family based training intervention programme for children with intellectual disabilities and less severe forms of challenging behaviour, the efficacy of which was previously established in a pilot project (Hudson et al., 2003). Although longitudinal data on effectiveness are required, programmes which offer intervention before behaviours become more severe and established clearly offer potentially important evidence for the effectiveness of secondary prevention. Given that parental stress and child challenging behaviours can have a negative reciprocal influence on each other (Hastings et al., 2006; Herring et al., 2006), these demonstrations are critically important. Tonge et al. (2006) were able to demonstrate in a randomized controlled trial that parental education and skills training produced improvements in parental mental health and family functioning that persisted for two years. There is also an emerging literature on resilience in families with an intellectually disabled member (e.g. Bayat, 2007; Broberg et al., in press), and the prospect that effective parental support programmes will contribute to its development.

Parallel interventions may be possible for the early amelioration of emerging mental health difficulties. The last three decades have seen a growing interest in the development of standardized procedures for assessing mental health needs in children and adults who have intellectual disabilities (e.g., Reiss, 1988; Reiss & Valenti-Hein, 1994; Moss et al., 1998), and these may have the potential to be used as screening tools for the identification of early difficulties.
Successful secondary prevention strategies for mental health difficulties in the non-learning disabled population involve multi-modal interventions that mirror the principles of much of the work described above. Intervention often takes place at the child, family and school level and may involve, for example, cognitive-behavioural interventions that help build emotional resilience, problem-solving and self-confidence, thereby reducing anxiety related problems; help to cope with major life events in order to ameliorate depressive reactions; interventions designed to foster family resilience; and school-based social skills training (WHO, 2004). Similar interventions, targeted at individuals showing early signs of emotional problems, could be equally helpful for children with intellectual disabilities.

**Tertiary Prevention**

Although the overall volume of available data remains comparatively small and is essentially dependent on single-case methodology, the strongest evidence base for intervention with challenging behaviour at the present time is derived from behaviour analytical approaches. Over the last two decades, a series of meta-analyses have repeatedly supported this position (Campbell, 2003; Didden et al., 1997, 2006; Marquis et al, 2000; Scotti et al., 1991, 1993; Whitaker, 1993). In keeping with the objectives of tertiary prevention, the central aim of the positive behavioural support approach referred to above is to achieve lifestyle changes for affected individuals. Thus, while reductions in the frequency, intensity or duration of challenging behaviours are a central objective, such changes are only important if they result in greater access to and participation in community settings, improved personal skills and relationships (Horner et al, 1990). Carr et al. (1999) summarised the strengths and weaknesses of the evidence base for this emerging approach by conducting a meta-analysis of 109 published articles that featured a total sample of 230 participants and 366 different interventions. The results indicated that 90% or more reductions in behaviour were achieved in 52% of
interventions, and 80% or more in 68% of interventions. Positive findings were also reported in relation to generalisation of change across settings and intervention mediators and successful maintenance of up to two years; only a minority of studies reported data on the latter however. Changes in levels of adaptive behaviour, effective lifestyle changes, and positive consumer evaluations were also evidenced, but were again only reported in a minority of studies. While this is reflective of the fact that this review was conducted at an early point in the use of this approach, it also demonstrates an evidence gap between its stated aspirations and the current evidence base. Further difficulties arise from the fact much of the historical evidence is derived from interventions conducted in controlled, clinical settings rather than integrated community locations and, to some extent, with less severe behaviours. There is evidence that both these trends are gradually changing however (Carr et al, 1999; Bambara et al., 2004; McClean et al., 2007) and this is an important consideration given that tertiary prevention strategies will, by definition, focus on more severe and chronic forms of challenging behaviour. Research suggests that this will involve targeting between 4-15% of people with intellectual disability (Borthwick-Duffy, 1994; Emerson, et al., 2001; Holden & Gitlesen, 2006; Lowe, et al., 2007a).

In keeping with the focus on both molar and molecular variables, contemporary behavioural interventions also place considerable emphasis on the importance of achieving change within the systems that typically mediate behavioural intervention (both in relation to family members and support staff) in order to maximise therapeutic impact. This has led, for example, to the development of the concept of ‘goodness of fit’ between behavioural interventions and mediating systems (Albin et al., 1996) and the development of quality assurance systems that are tailored to supporting behavioural intervention (LaVigna et al., 1994). There is some limited evidence to indicate that interventions that feature more systemic interventions are associated with better outcomes (Carr et al, 1999).

Given the fact that carers supporting individuals with challenging
behavioural or mental health needs are in themselves likely to experience strong emotional reactions and heightened stress levels (Bromley & Emerson, 1995; Cottle et al, 1995), there is some promising initial research (Singh et al, 2006, 2009; Noone & Hastings, in press) showing that the addition of mindfulness-based interventions with paid carers can enhance the impact of behavioural training and produce improved outcomes in terms of reduced rates of challenging behaviour, restraint and emergency medication use, and staff and service user injuries. While training in behavioural principles may therefore impact on attributions and subsequently increase helping behaviour, mindfulness approaches could help to reduce levels of expressed emotion (McGill et al., 2007; Lowe et al, 2007b ; Singh et al., 2006; Weigel et al, 2006). Gavidia-Payne & Hudson (2002) suggest that multi-modal interventions of this type are also likely to be helpful in family settings.

One of the defining characteristics of contemporary behavioural approaches is that they differentiate between proactive strategies for behaviour change and reactive strategies to safely manage behaviours that pose risks either to the person with intellectual disabilities or those who care for them. Despite evidence for the high use of intrusive procedures such as physical restraint, seclusion and emergency medication with this population, the evidence base for their effectiveness in helping safely manage severe challenging behaviours is conspicuous by its absence (Allen, 2001; McDonnell, 2009). This is a significant concern given the inherent physical and psychological risks associated with the use of these procedures (Allen, 2008; Valdovinos et al, 2005). Importantly, there are an increasing number of accounts of the efficacy of restraint reduction procedures in human services (Thompson et al, 2008; Carter et al, 2008) and a number of specific demonstrations within service for people with intellectual disability (Sturmey & McGynn, 2003; Saunders, 2009).

Paclawskyj & Yoo (2004) review how behavioural methods such as systematic desensitization, shaping, relaxation training, graduated exposure, response prevention, differential reinforcement and social skills training have
been successfully used in the treatment of a variety of mental health problems in people with intellectual disability. There is also an increasing interest and emerging evidence base in relation to other psychological therapies in this field. Cognitive behavioural approaches, and particularly interventions for anger control, appear to offer promise with individuals with milder intellectual disability (Lindsay et al, 2004; Willner et al, 2002); there is also evidence for the effective treatment of depression using cognitive methods (McCabe et al, 2006). A meta-analysis by Prout & Nowak-Drabik (2003) concluded that broader psychotherapeutic approaches can also have beneficial effects, although the validity of this conclusion has been subject to severe criticism (Sturmey, 2005) and counter criticism (Beail, 2005; Hurley, 2005). A more recent review by Parkes & Hollins (2007) stated that there is some evidence that the use of psychodynamic therapies have the potential to be of benefit in reducing both general psychological distress and specific forms of challenging behaviour, such as aggression. Similarly, Dagnan (2007) indicates that, while the evidence base for cognitive behavioural approaches for mental health and behavioural difficulties within this population is comparatively weak, the existent data offer sufficient promise to suggest their use. By definition however, their effectiveness is likely to be limited in people who have more severe deficits in their verbal and cognitive skills.

Despite their frequency of use (Fleming et al., 1996; Kiernan et al., 1995; Robertson et al., 2000), there is also a ‘relative absence of data’ (King, 2007) for the efficacy of psychopharmacological interventions for mental health and behavioural difficulties in people with intellectual disabilities. The latter author reviews current evidence for the impact of medication in treating a variety of conditions (including anxiety disorder, mood disorders, psychosis and attention deficit disorder) in this population. The treatment of behavioural disorders via medication is particularly controversial. Tyrer et al., (2008) recently showed that there was no difference between antipsychotic medication (haloperidol and risperidone) and placebo in effect on non-specific aggressive behaviour, and a related study by Romeo et al., (2009) concluded
that the treatment of challenging behaviour with antipsychotics is not cost-effective. Ahmed et al. (2000) also reported that it was possible to withdraw antipsychotic medication in one third of participants and to reduce them by half in another fifth without any subsequent escalation in challenging behaviour. Deb et al., (2006) have produced detailed practice guidelines for the use of psychotropics as a component of comprehensive intervention for challenging behaviours.

Studies of this type appear to confirm the perspective that it is the underlying causes of challenging behaviour and emotional difficulties that should be the focus of intervention rather than the treatment of isolated behaviours (Gardner et al., 2001). Medication does however have a central role in the treatment of properly diagnosed drug-responsive psychiatric disorders and there is a series of good practice guidelines available that should govern their use for this purpose (Reiss & Aman, 1998; Rusch & Allen, 2000).

The distinction between challenging behaviours and mental ill-health has sometimes been considered as a false dichotomy (Croce, 2009; Wieseler & Hanson, 1999) and one that may foster incomplete or inappropriate interventions. King (2007) suggests that behavioural and diagnostic formulations are not mutually exclusive however and can be viewed as acting in a complementary fashion rather than in opposition. Thus, when either challenging behaviours or psychological distress is apparent, a comprehensive, multi-modal case formulation is required that embraces organic, genetic, psychological and environmental factors (Croce, 2009; Gardner, 2002; Dosen 2005a, b; Sturmey, 2007). Integrated assessment needs to be followed by integrated intervention that aims to re-establish the health and well-being of the person concerned. There are a number of excellent theoretical accounts for how such an approach should work in practice (e.g., Thompson et al, 1994; Wieseler & Hanson, 1999), although practical demonstrations are very limited in number (Lowry & Sovner, 1992; Sovner et al., 1993).
As with positive behavioural support, a number of authors (Szymanski & King, 1999; Volkmar et al., 1999) have argued that the treatment in persons with intellectual disability and mental health problems should be directed not merely to remove the symptoms of the disorder, but toward helping the person to achieve the highest attainable level of quality of life.

**Summary Model**

The overall model proposed in this paper is summarised in figure 1. This reflects the three levels of prevention described above which specific examples drawn from the text. It also reflects the fact that reducing numbers of people with intellectual disability will be targeted as the focus switches from primary to secondary and tertiary prevention, but that the intensity of intervention will increase and the potential benefits of intervention may decrease across these intervention tiers.

Whilst secondary and tertiary interventions targeted at high-risk groups are likely to produce the best results for the individuals concerned, optimum outcomes for the population are more likely to come from universal primary prevention interventions as they aim to change the distribution of risk in that population. Rose (1992) refers to this as the ‘Prevention Paradox’. The different tiers of intervention preventions are therefore complementary rather than alternate courses of action.

Insert figure 1 about here.

The percentages of people requiring intervention at secondary and tertiary levels reflect the epidemiological data referred to above.

**Discussion**
A significant proportion of people with intellectual and developmental disability present with emotional and behavioural problems. The current prevailing strategy is one of ‘diagnose and treat’, whereby an individual receives access to intervention once the behaviour or emotional problem is firmly established in their behavioural repertoire, and by which point considerable cost has been incurred (in terms of the quality of life of the person, their family and in terms of the financial implications for service commissioners) and treatments are less likely to be effective. Even then, the numbers receiving intervention may not reflect numbers in need (Oliver et al, 1987; Harris & Russell, 1989; Lowe et al, 2007a; Qureshi, 1994).

In this paper, an alternative strategy has been outlined- one that emphasises a reduction in the incidence of behavioural and emotional difficulties through the use of primary and secondary prevention strategies. This approach holds many benefits over a ‘diagnose and treat’ model, potentially reducing the overall cost incurred by challenging behaviour over the lifespan and freeing up resources for better quality tertiary prevention for those who continue to present with severe emotional and behavioural difficulties.

While an ‘absence of evidence’ should not be mistaken for ‘evidence of absence’ (WHO, 2005), the model described has been primarily conceptual in nature. At present, the most substantial evidence base concerns tertiary prevention, something which in itself reinforces a ‘diagnose and treat’ dynamic and works against the adoption of a more expansive model. Even then, existent evidence is limited and relates much more to behavioural difficulties rather than emotional and psychological health in its broadest sense. Research that demonstrates how the development of the promise of this preventative approach can be translated into practice and impact on both the behavioural and emotional needs of people with intellectual disability is therefore a sine qua non; although the potential research topics here are many and varied, the priorities are likely to include:
1. Greater attention to the theory, concepts and methodology from the study of developmental psychopathology in the general population. There has been real growth in this area (from which the intellectual disability research community is almost completely detached).

2. Testing out intervention effectiveness (i.e., whether it works in practice) and efficiency (i.e., whether it is worth doing), rather than efficacy (i.e., whether it does more harm than good under ideal circumstances) (Haynes, 1999). At present, the case for investment is significantly weakened by the absence of sufficient, credible policy relevant evidence for any specific intervention approach.

3. High-quality, longitudinal research on primary and secondary prevention in early childhood.

4. Research that demonstrates intervention impact in terms of, for example, improving quality of life as well as changes in behavioural or mental health status.

5. Research that addresses policy and practice issues related to scaling up the provision of and access to evidence-based interventions, especially in low and middle-income countries.

In addition to robust empirical research, there is also scope for more descriptive longitudinal studies which are primarily observational in nature and which aim, for example, to track the impact of policy changes via repeated surveys and routinely collected data in health and social care services (Sturm, 1999).

The above priorities also carry implications for the role of organisations such as the International Association for the Scientific Study of Intellectual
Disabilities (IASSID) in supporting both high-quality research and the global development of effective social policies. Both outcomes require an approach in which work on intellectual disability is much better integrated within broader agendas. In social policy terms, for example, we have seen international attention to the development of effective parent training procedures without the full inclusion of families of children with intellectual disabilities. Similarly, the kind of research required is likely to necessitate international collaborations that attend to the development of preventative strategies that are effective across a wide range of circumstances.

Research and practice in intellectual disability has always been open to the criticism of parochialism. In a global world we must now attach ourselves much more to the mainstream as we seek inclusion not just for those we support, but also for ourselves.
References


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treatment and follow-up of consecutive referrals and a waiting list comparison. Clinical Psychology & Psychotherapy, 11, 4, 255-264.


Figure Caption:

Figure 1: The preventative model
Tertiary prevention:
4-15% of population. Intensive behavioural and psychiatric intervention.

Secondary prevention:
Up to 30% of population focused on all those with known risk factors (male, severe-profound ID, communication difficulties, mobility problems, presence of genetic markers) and followed-up with behavioural & psycho-social early intervention.

Primary prevention:
Universal, selective and indicated interventions that include poverty reduction, reducing health discrepancies, providing service environments with wide-spread competence, reducing in utero exposure to known toxins etc.

‘Changing the odds’

‘Beating the odds’

Potential impact of intervention

Required intensity of intervention

Numbers requiring intervention