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# Self-injurious behaviour in people with intellectual disability and autism spectrum disorder

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## **Abstract**

**Purpose of review:** This review summarises the recent trends in research in the field of self-injurious behaviour in people with intellectual disability and autism spectrum disorder.

**Recent findings:** New data on incidence, persistence and severity add to studies of prevalence to indicate the large scale of the clinical need. A number of person characteristics have been repeatedly identified in prevalence and cohort studies that: 1) can be considered as risk markers (e.g. stereotyped behaviour, autism spectrum disorder) and 2) indicate possible causal mechanisms (e.g. sleep disorder, anxiety). Studies have started to integrate traditional operant learning paradigms with known person characteristics and reviews and meta-analyses of applied behaviour analytic procedures can now inform practice.

**Summary:** Despite these positive developments interventions and appropriate support falls far short of the required need. Expansions in applied research are warranted to develop and evaluate innovative service delivery models that can translate knowledge of risk markers and operant learning paradigms into widespread, low cost routine clinical practice. Alongside this, further pure research is needed to elucidate the direction of causality of implicated risk factors, in order to understand and intervene more effectively in self-injury.

**Keywords:** self-injury; intellectual disability; autism spectrum disorder; applied behaviour analysis

## **Introduction**

Self-injurious behaviour in children and adults with intellectual disability and/or autism spectrum disorder remains a significant challenge for clinicians and carers. The research field has been characterised by fragmentation with little evidence of integrated biopsychosocial models of cause and hence intervention. However, there are indications that researchers are drawing on previously separate literatures and concepts, with research methods broadening in scope to capture data that reflect the possibility of different causes across and within individuals. Additionally, there are now studies of incidence, persistence, risk markers and the translation of risk markers into risk factors alongside the emergence of a strong literature on pain as a potential cause of self-injury. These trends are welcome and have clear implications for both preventative and responsive intervention strategies. In this review we highlight recent studies that provide novel data or insights into a broader and more nuanced understanding of self-injurious behaviour and draw out the implications for research and practice.

## **Incidence, Prevalence and Persistence**

Whilst there have been numerous studies of the prevalence of self-injurious behaviour, total population data on incidence, persistence and severity are scarce. In a total population study of approximately a thousand children with severe intellectual disability the prevalence of the most severe self-injury was estimated to lie between 4% and 5% [1\*]. In this study severe self-injury was defined as occurring frequently and as very difficult to manage, suggesting this prevalence estimate captures the population in most need of services and support. Interestingly, where there were data for both teacher and parent ratings of severity for the same child, parents were significantly more likely to rate the behaviour as more severe (approximately 5% vs. 19% respectively) suggesting prevalence estimates need careful interpretation.

The first published study of incidence in children with severe intellectual disability [2\*] estimates cumulative incidence over one year at 4.7% (compared to aggression and destruction at 8.45% and 8.23% respectively) and persistence at 58%. Importantly, onset of self-injury was predicted by a different behavioural marker (repetitive behaviour) to that which predicted the onset of aggression (impulsivity). In a similar study cumulative incidence for self-injury in autism spectrum disorder (ASD) was estimated to be approximately 6% over one year with 78% persistence over three years [3\*]. In combination these data show that self-injury is still very much in evidence in these populations at a clinically significant level and the behaviour is persistent.

## **Person characteristics**

A sufficient number of robust prevalence and cohort studies have now been published to enable analyses of the relationship between the presence, severity and trajectory of self-injury and person characteristics (both stable and transient) to be conducted. Dependent on their relationship with self-injury, these characteristics might be identified at least as risk markers (associating with onset or trajectory) or risk factors (predicting but also causally implicated in self-injury). Examples of clear risk markers are severity of intellectual disability and the presence of autism spectrum disorder. An example of a possible risk factor is compromised health (leading to unidentified pain and hence self-injury). In delineating risk markers and factors there needs to be care in the identification of the characteristics of interest and the interpretation of their involvement in the aetiology and maintenance of self-injury. As this body of data comes together it has the potential to inform and challenge prevailing causal models and this trend is evident in recent studies.

It is perhaps surprising that behaviours such as self-injury and stereotypies have rarely been compared directly in children of typical development and those with intellectual disability (ID). In pre-schoolers both of these behaviours were more common in an ID group [4]. A recurrent finding in previous studies is that within the group of people with ID self-injury is more common in those who also have a diagnosis of ASD. This association was confirmed in those with profound and severe ID in a study of 370 adults that also identified higher levels of anxiety, stereotyped behaviour and impulsivity in those showing self-injury [5]. These person characteristics feature repeatedly in other studies [2\*,3\* and see below]. Similarly, in a study of 152 children with ASD, the severity of ASD was associated with the presence of self-injury [6].

Clearly the relationship between self-injury and ASD should be assessed carefully as many assessments of ASD include self-injury as a feature and there is significant shared variance conferred by the associations with intellectual disability. Additionally, some aspects of ASD may prove to be more important with regard to self-injury than others. In three studies [2\*, 3\*, 7\*] stereotyped behaviours predicted self-injurious behaviour and this relationship appears robust in the absence of a diagnosis of ASD. Interestingly, it has also been argued that a relationship between self-injury and stereotyped behaviour may be evident for a subgroup of those showing particular forms of self-injury but not others [8\*]. This approach to clustering person characteristics and carefully defining topography and severity of self-injury may prove to be helpful in identifying meaningful subgroups.

Finally, specific genetic disorders are firmly established as an important person characteristic associated with self-injury. However, less attention is paid to those syndromes that despite being associated with numerous risk markers, such as degree of intellectual disability, evidence significantly lower levels of self-injury than might be expected. Angelman syndrome is one example and a study of Rett syndrome described a similar comparatively low level [9]. The progression of work on risk factors for self-injury would be significantly enhanced by an evaluation of potential protective mechanisms in these syndromes.

The studies described above have identified possible risk markers. In other cohort studies associations between self-injury and person characteristics allude to causal factors that could prove influential with a number of recurrent themes. An interesting study of seemingly diverse behaviours and characteristics in 109 children and adolescents demonstrated a relationship between sleep disorder, self-injury and anxiety [10\*] and anxiety is related to some forms of repetitive behaviour in fragile X syndrome [11]. Low interest and pleasure have been shown to predict self-injury at follow-up [12] and sleep disorder has also been associated with challenging behaviour more generally in people with profound intellectual disability [13]. These associations between internal states and self-injury are interesting but often difficult to interpret, particularly with regard to direction of causality. There are clear methodological challenges in assessing anxiety for example but there are emerging research paradigms that can be employed. Higher cortisol levels, for example, are evident in children with ASD who showed higher levels of stereotyped behaviour [14\*]. In combination, these studies indicate a need for robust assessments of internal states to accurately identify anxiety because there is a strong association between stereotyped behaviour and self-injury and emerging evidence of an association between self-injury and anxiety (both by implication and more directly). Causal models of self-injury may need to account for this association with anxiety (either as driver or passenger) and the same may be argued for sleep disorder.

Over the last decade a number of authors have suggested pain might be important as a cause of self-injury but the investigation of pain has proved problematic. The assessment of the presence of pain often relies on self-report which is frequently compromised in those at the highest risk of self-injury. However, there are important and innovative studies that are showing the way ahead. The work by Symons is notable as it shows an intriguing relationship between self-injury and high epidermal nerve fibre density values in a small sample of children [15, 16\*\*]. This finding may help explain the emerging data on a positive relationship between self-injury and sensory disorders in people with ASD and is promising. In the same vein a study of the relationship between behavioural correlates of pain, assessed by an informant measure, and self-injury in 51 children showed higher

scores on the informant measure in the children showing self-injury than those who were not [17\*\*]. Inevitably there is a question about causal direction in studies of this kind but the evidence is growing that pain should be considered as a possible cause of self-injury in clinical services and perhaps as the first priority.

### **Assessment and intervention**

A search for recent studies on assessment and intervention for self-injury revealed a list of studies characterised by demonstrations of operant learning paradigms in applied settings using single case experimental designs with varying degrees of strength of design and method and a paucity of psychopharmacological studies that focus on self-injury specifically. This result perhaps reflects the maturity of the applied behaviour analytic field as many methods are now standard clinical practice and the absence of psychopharmacological studies is perhaps evidence of the limited understanding of potential pharmacological targets in causal models of self-injury.

Other evidence of the maturity of the applied behaviour analytic field is the emergence of datasets on types of interventions that allow analysis of effectiveness alongside other variables. In each case the intervention has been effectively demonstrated in numerous single case experimental designs with attention to operant principles that might enhance effectiveness. A review of Functional Communication Training demonstrates that schedule-thinning with the use of discriminative stimuli can maintain effectiveness [18\*] and similarly a meta-analysis of studies on Noncontingent Reinforcement (NCR) studies provides evidence of strong effects again when combined with schedule thinning [19\*\*]. The latter study is important as the procedure of NCR is comparatively straightforward to implement and maintain.

Other developments in the operant literature are the refinement of methods of functional analysis, for example when identification of function is ambiguous [20] and the identification of patterns of responding for automatically reinforced self-injury that might indicate different causes [21\*]. These studies extend the use of functional analysis. A welcome addition to the literature are studies that combine person characteristics with operant analyses to identify possible phenotype x environment interactions, for example escape behaviour in fragile X syndrome [22]. Additionally, more transient states such as fatigue have been demonstrated to interact with environmental contingencies to drive self-injury [23\*\*]. This is of interest given the possible relationship between sleep disorder and self-injury [see above, 13].

As the literature on functional analysis extends in these ways and specific methods of functional analysis become established by repeated use in studies it is important to continue to assess psychometric properties of different methods that have different ethical constraints and demands on resources. Experimental and descriptive functional analysis and informant questionnaire assessments yield different results for social functions of behaviour but agreement on automatic reinforcement is higher [24]. This finding has been described previously and suggests there is still some way to go to optimise the use of functional analysis. A welcome addition to the literature is the consideration of how safe the use of functional analysis is in practice. The results of a review of 99 cases showed that injury rates were ‘relatively low’ [25].

The studies reviewed suggest that methods of assessment such as functional analysis and the applied behaviour analytic approach more generally are considered as the assessments and interventions of choice. This view has been adopted in the relentless stream of policy documents and guidelines that are issued regularly. It is sobering then to consider the results of two empirical studies of clinical practice. The first showed that of 205 practitioners the majority used descriptive methods of functional analysis only. Additionally, whilst 67% of respondents indicated that they thought functional analysis was useful, only 37% used the results to inform interventions [26]. This finding is consistent with audits of positive behavioural support packages that frequently cite applied behaviour analysis as informing intervention but rarely implement it in practice. The second study is a stark reminder of the challenge of service delivery. For children who show clinically significant levels of challenging behaviour (high frequency and very difficult to manage) the odds ratio for the need for behavioural intervention identified by teachers was 13 times that for children not showing these behaviours. However, the odds ratio of children showing these behaviours having relevant clinical contact was only twice that of children not showing these behaviours. The identified need for behavioural intervention was significantly higher for primary carers than teachers and the most likely professional contact was with a social worker [1\*].

## **Conclusions**

Incidence and persistence data have added to our knowledge of the scale of the clinical problem of self-injurious behaviour and prevalence data appear to indicate that self-injury remains a significant challenge. In some respects there is cause for optimism. The data on person characteristics can inform risk driven prevention and early intervention strategies. Additionally, these characteristics warrant investigation to understand the nature of their relationship with self-injury. Impulsivity, stereotyped behaviour, anxiety and sleep disorder are each promising candidates to help extend causal models of self-injury. The developments in methods for identifying

pain are encouraging and studies of the relationship between health, pain, sleep and self-injury are warranted. The role of impulsivity and repetitive behaviours might lead to casual models including compromised behavioural inhibition as explanatory of phenomena such as preferred imposed restraint and self-restraint. The role of anxiety is likely to prove important and research might usefully focus on whether episodes of anxiety evoke self-injury or are the result of self-injury that cannot be easily inhibited. Investigations in these areas are likely to be productive and can inform interventions strategies.

With regard to applied behaviour analysis and intervention delivery the challenges are different and related to implementation as opposed to discovery. Demonstrations of the reduction of severe self-injury using operant methods were first described 50 years ago. It is not clear why there has been such widespread failure (in the UK at least) to make these kind of effective interventions available and it is also questionable that existing services are likely to be able to rise to the challenge. There is merit in pursuing applied research that identifies causes of self-injury that can be effectively treated with existing interventions. Pain is the best example. However, there is also clearly a need to optimise assessment protocols, increase the number and availability of appropriately trained practitioners and develop service delivery strategies that match the level of unmet need.

## **Key points**

The prevalence of severe self-injury in children with severe intellectual disability is approximately 5%, incidence is approximately 5% and persistence is approximately 60%.

Behavioural risk markers for self-injury include stereotyped behaviour and impulsivity.

Correlates of self-injury, such as sleep disorder and anxiety, warrant further examination.

Service and intervention provision for self-injury falls significantly below the level of clinical need.

## **Acknowledgements**

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Papers of particular interest published within the annual period of review have been highlighted as:

\* of special interest

\*\* of outstanding interest

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