

Self-injurious behaviour in people with intellectual disability

Frank J. Symons, Darragh P. Devine and Chris
Oliver

*Cerebra Centre for Neurodevelopmental Disorders,
School of Psychology,
University of Birmingham*

Please use this reference when citing this work:

Symons, F. S., Devine, D. D. and Oliver, C. (In press). Self-injurious behaviour in people with intellectual disability. *Journal of Intellectual Disability Research*.

It is approaching 50 years since the first empirical studies of self-injurious behaviour (SIB) in people with intellectual disability and autism spectrum disorder were conducted by Ivar Lovaas and colleagues (Lovaas, Freitag, Gold et al., 1965; Lovaas and Simmons, 1969). These early studies generated significant clinical and research interest as they demonstrated the potential for applied science, via the principles and methods of operant learning theory, to contribute to the understanding and reduction of one of the most distressing and intractable problems in the field of intellectual disability. As these principles and methods were translated into the technology of Applied Behaviour Analysis, different classes of reinforcement of self-injury were identified and the seminal reviews of the resultant research by Bachman (1972) and Carr (1977) began to shape the research agenda that is still evident today.

The review by the late, and sorely missed, Ted Carr was influential in a number of ways. First, it outlined a behavioural taxonomy based on the class of reinforcement and its social or sensory delivery. Second, the descriptions of the evidence, derived from manipulations of contingencies, provided the framework for experimental functional analytic assessment methods later developed by Iwata et al. (1982) and Carr and Durand (1985) amongst others. Third, Carr suggested the causes of self-injury might have an operant basis and/or a biological cause. In doing so he identified and tried to amalgamate the polarised positions adopted in the field. Carr did two other things that remain influential today. He promoted both psychological and biological research into the causes of self-injury and he made explicit the basis of a clinical assessment framework that encapsulated both individual characteristics and manipulation of environmental variables. In this special edition of JIDR we have brought together a series of empirical papers and reviews that cover the range of contemporary research on self-injury identified in Carr's review. By doing so, we can evaluate progress toward a more complete account of self-injury that might lead to an effective, comprehensive, data driven clinical assessment.

The need for research that spans environmental and biological variables is evident from a critical appraisal of whether operant learning can offer a complete account of self-injurious behaviour. There are a number of well established observations that are pertinent. The documented association between some genetic disorders and self-injury and the difference in forms of self-injury between syndromes (Arron et al., 2011) alludes

to causal variables that are either not in the environment or are operant in nature but interact with aspects of the behavioural phenotype associated with a genetic syndrome (Langthorne and McGill, 2008; Tunncliffe and Oliver, 2011). A similar argument may be made for the growing evidence that some psychological characteristics, such as repetitive behaviour, autism spectrum disorder and impulsivity are associated with self-injury and, in the case of repetitive behaviour, more severe self-injury (Arron et al., 2011; Oliver et al., in press). In this edition, Richards et al. (2012) extend these observations to show that ASD characteristics in those who have Fragile X or Down syndrome are associated with a higher prevalence of self-injury and that within the high risk group of those with ASD, impulsivity is associated with self-injury. Clearly these findings suggest a complete account of self-injury should identify why these individual characteristics are observed in those showing the behaviour. The review by Biswas and Furniss (2012) in this edition updates the literature on these characteristics and highlights the implications for intervention.

At about the time that Lovaas published the first operant accounts of self-injury in humans, early reports of self-injury in animals appeared. These reports described pharmacologically-induced self-biting in rats and mice (Genovese et al., 1969; Peters, 1967), and spontaneous self-biting in rhesus macaques that were raised in impoverished conditions (Harlow and Harlow, 1962). Further investigations using these models have helped to reveal neurobiological abnormalities that may contribute to the aetiology of self-injury. For example, cortical lesions enhanced the ability of pemoline to induce stereotypy and self-biting behaviours in rats (Cromwell et al., 1998), demonstrating potential links between the pharmacological model and cortical dysfunction that is common in those with intellectual disability. Harlow's self-injurious monkeys exhibited profound levels of stereotypy during and after their social isolation (Harlow and Harlow, 1962; Harlow et al., 1965), and it was eventually found that these animals had significant alterations in striatal chemoarchitecture (Martin et al., 1991). These effects of early impoverishment are redolent of effects in human children. When severely environmentally-deprived children who were raised in Romanian orphanages were assessed in the 1990s, 47% were reported to engage in rhythmic stereotypy and 24% in self-injurious behaviour (Beckett et al., 2002). Indeed, there appears to be a high degree of co-morbidity between stereotypy and self-injury in people with intellectual disability (Gal et al., 2009; Oliver et al., in press), and abnormalities in striatal function are thought

to be an important component in both behaviours (Turner & Lewis, 2002). In this edition, Muehlmann and Lewis (2012) review the evidence for shared phenomenology and pathophysiology, drawing upon evidence from a combination of human conditions and animal models.

Over the years, the biological basis of self-injury has been most extensively characterised in a neonatal 6-hydroxydopamine lesion model (Breese et al., 1984, 2005). Additional investigations have been reported with a variety of pharmacological manipulations including caffeine, pemoline, and Bay K 8644 administration (Devine, 2011). Evidence is also building that specific transgenic manipulations may promote expression of spontaneous (Welch et al., 2007), or pharmacologically-induced (Keebaugh et al., 2011) self-injury in laboratory mice. Taken together, these studies provide evidence that abnormal neurochemical signalling contributes to the pathophysiology of self-injury. Most investigations have focused upon striatal dysfunction, especially in dopaminergic neurotransmission. However, the specific neurochemical substrates are subject to ongoing investigation, and there is clearly a need for broader assessment of hormones, neurotransmitters, and cellular signalling mechanisms. In one recent study, it was reported that individual differences in stress responsiveness confer individual differences in vulnerability for pemoline-induced self-injury in outbred rats (Muehlmann et al., 2011). In this edition, these investigators extend those observations, and report that pre-exposure to social/emotional stressors enhances vulnerability for pharmacologically-induced self-injury (Muehlmann et al., 2012). Taken together, the studies indicate important roles for stress-responses in the aetiology of self-injurious behaviour, and suggest that stressful situations should be explored further as potential establishing operations for expression of SIB.

Evidence has also accumulated that self-injurious behaviour occurs in animals under a broad variety of conditions outside of the laboratory setting. This includes domestic pets (Jenkins, 2001; Schwartz, 2003), farm animals (Dodman et al., 1994) and zoo specimens (Novak et al., 2002). Self-injury was even reported in the case of a wild Gombe chimpanzee during a period of severe distress (Goodall, 1986) (although it should be noted that self-injury appears to be uncommon in wild animals; Dellinger-Ness and Handler, 2006). These observations suggest that the propensity to self-injure is broadly present in vertebrate species. Thus, animal models of self-injury appear to represent

neurobiological perturbations that contribute to vulnerability for self-injury, or they imbue setting conditions that may promote the ongoing expression of self-injurious behaviour. Overall, these models share interesting parallels with self-injury in human disorders. An important challenge for future investigations is to identify the ways in which biological predispositions and environmental stimuli interact to contribute to the aetiology and expression of self-injury in animal models of this debilitating behaviour.

The work by Courtemanche et al. (2012) and Peebles et al. (2012) highlights a bio-behavioural focus on the potential role for pain and pain perception in emerging models of self-injury informed from the 'bench' as well as by practice. Under normal circumstances pain is protective, it is the body's signal that something is wrong, and can be related to acute or chronic medical conditions. It has been recognized for some time that undiagnosed medical conditions that could be expected to be painful co-vary with SIB (see an early paper on this by Bosch et al., 1997 as well as a review by Kennedy and O'Reilly, 2006). What is less clear is whether the neurobiological mechanisms regulating nociception may be 'co-opted' by the repeated tissue damage associated with chronic SIB. From this perspective, it may be relevant to consider more directly the physiological sensory mechanisms relevant to pain (and possibly itch) transmission and regulation in relation to chronic SIB (see Edelson, 1984, for an earlier account). Consider, for example, that peripheral nerve damage secondary to viral infection (e.g., post-herpetic neuralgia) can lead to neuropathic itch (so severe, in some cases that affected individuals scratch to the bone and in a few notable case through bone as well) among otherwise healthy individuals with no psychiatric history (Oaklander, Cohen, & Raju, 2002). Histological evidence from skin samples implicates abnormal peripheral innervation of primary sensory afferents. Preclinical rodent models, developed to examine putative central nociceptive mechanisms underlying pathological itch/self-injurious scratch, also implicate abnormal innervation as well as local and central immune response (Brewer, Lee, Downs, Oaklander, & Yesierski, 2009). Such findings underscore recent observations in skin biopsies taken from small samples of individuals with chronic SIB and intellectual disability showing altered peripheral epidermal innervation densities and corresponding elevations in concentrations of substance P (SP) in the skin of individuals with SIB, sampled from non-self-injurious body sites (Symons, 2011). Intriguingly, in many samples, extensive mast cell degranulation (consistent with immune mediated inflammatory

response) was also observed. These findings point to the possible relevance for a ‘peripheral biomarker’ approach to improve our understanding of the pathophysiology associated with SIB, or more realistically SIB subtypes. However, the real value here would be in knowing whether the peripheral biomarkers were causally related to SIB and had any predictive value in understanding treatment response; either pharmacological or behavioural.

The empirical evidence and critical reviews presented in this special edition clearly indicate the need for models of self-injury that can account for all established observations of influential psychological, environmental and biological variables. Hence, the need for a broad research agenda (and funding for that agenda) remains paramount. However, empirical studies are likely to be more productive if they are open to the methods and concepts of other perspectives. This is nicely demonstrated in this edition in the study by Courtemanche et al. (2012) which used lag sequential analysis, traditionally employed in operant research, to examine possible pain related behaviour as a precursor to self-injury. Similarly, the innovative review by Peebles et al. (2012) indicates how pain perception might moderate the relationship between self-injury and social reinforcement.

The future directions for research that are likely to improve assessment and intervention in clinical practice include a valid and reliable technology for pain assessment (both indicators of presence of pain and compromised pain perception), the effective delivery of applied behaviour analysis at the point of service access, and evaluation of person and environmental characteristics that might be translated into risk markers for the future development of clinically significant self-injury. These clinical directions should be informed by expanding basic research in the potential interactions between operant learning theory, psychological characteristics, and neurobiological mechanisms that encode the pathophysiological basis of syndromic and idiopathic self-injury.

The greatest immediate challenge is to increase the availability of demonstrably effective intervention for self-injury shown to be socially reinforced and amenable to reduction using the methods of applied behaviour analysis. The inability of services to make these interventions routinely available and affordable, or only available to those with specific diagnoses, seems driven by a proclivity for the use of treatments for which there is little, if any, evidence combined with the lack of appropriately trained professionals. At the

same time, we do not have a broad enough empirical base regarding implementation and adherence regarding the successful ‘uptake’ of effective interventions. Despite a few notable longer-term intervention studies (by Wacker and colleagues; e.g., Steege, et al., 1998), most intervention studies are focussed on establishing efficacy or explicating a mechanism. These remain necessary agendas but so does work addressing family and related care systems in relation to ‘diffusion of innovation’ to increase our understanding of the variables related to successful intervention implementation that actually maintains and reduces the *burden* of self-injury. In this respect, the pervasive, enduring and widespread failure of clinical psychology training to lead on these issues is lamentable. The stark contrast between the mandatory requirement for training of clinical psychologists in cognitive behavioural methods and the neglect of applied behaviour analysis warrants action, not least because of the inadvertent bias toward training in interventions that exclude those with the most severe disability.

By publishing this special edition we hope to reinvigorate interest in self-injury and promote research that can be translated into effective intervention. In this editorial we wanted to acknowledge the legacy of Ted Carr of promoting the systematic evaluation of the potential causes of this behaviour from different perspectives. It is this approach that will ultimately prove beneficial to those who show self-injury and those who are charged with responsibility for delivering effective interventions at the point of need.

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