

# **HHS Public Access**

J Intellect Disabil Res. Author manuscript; available in PMC 2019 December 01.

Published in final edited form as:

Author manuscript

J Intellect Disabil Res. 2018 December ; 62(12): 1108–1113. doi:10.1111/jir.12430.

# Classification of Self-Injurious Behaviour Across the Continuum of Relative Environmental-Biological Influence

Louis P. Hagopian<sup>1,2,3</sup> and Michelle A. Frank-Crawford<sup>1,3</sup>

<sup>1</sup>Department of Behavioral Psychology, Kennedy Krieger Institute

<sup>2</sup>Department of Psychiatry and Behavioral Sciences, Johns Hopkins University School of Medicine

<sup>3</sup>University of Maryland, Baltimore County

## Abstract

Self-injurious behaviour (SIB) is generally considered to be the product of interactions between dysfunction stemming from the primary developmental disability and experiences that occasion and reinforce SIB. As a result of these complex interactions, SIB presents as a heterogeneous problem. Recent research delineating subtypes of SIB that are non-socially mediated, including one that is amenable to change and one that is highly invariant, enables classification of SIB across a broader continuum of relative environmental-biological influence. Directly examining how the functional classes of SIB differ has the potential to structure research, will improve our understanding this problem, and lead to more targeted behavioural and pharmacological interventions. Recognizing that SIB is not a single entity, but is comprised of distinct functional classes would better align research with conceptual models that view SIB as the product of interactions between environmental and biological variables.

#### Keywords

automatic reinforcement; biological variables; environmental variables; self-injurious behaviour

Self-injurious behaviour (SIB) is generally considered to be the product of interactions between dysfunction stemming from developmental disabilities and experiences that occasion and reinforce SIB (Furniss & Biswas, 2012; Oliver et al., 2013; Richman & Lindauer, 2006). Given the variability and complexity of these interactions, SIB is likely to have multiple etiologies. This might also explain why it is such a heterogeneous phenomenon that varies across numerous dimensions, including its reinforcing function, co-occurrence with other problem behaviours, and resistance to treatment. Despite general recognition that environmental-biological interactions lead to the emergence and maintenance of SIB, research on SIB rarely examines these interactions. Behaviour analytic studies typically identify the reinforcing function of SIB and other dimensions relevant to clinical care, but rarely examine differences across functional classes or the biology of SIB.

Address correspondence to: Louis P. Hagopian, Department of Behavioral Psychology, Kennedy Krieger Institute, 707 N. Broadway, Baltimore, MD, 21205. Phone: 443-923-2841. hagopian@kennedykrieger.org.

Research on the biology and phenomenology of SIB and medication studies carefully characterize participants (e.g., according to age, gender, disability), but these studies rarely examine the function of SIB (few exceptions exist; e.g., Garcia & Smith, 1999).

How SIB changes across different environmental contexts can inform us about the relative contribution of environmental and biological variables in its maintenance. Decades of behavioural research using functional analysis indicate SIB is maintained by social variables (e.g., access to attention or escape) in approximately two-thirds of cases (Beavers, Iwata, & Lerman, 2013). In approximately one-quarter of cases however, SIB occurs and persists independent of social variables (in a small percentage of cases, the function cannot be determined). The term automatic reinforcement is used to describe this category because it is thought that SIB itself produces some type of reinforcement (LeBlanc, Patel, & Carr, 2000). A biological "reinforcement" process has been posited by many researchers (e.g., release of endogenous opioids, and pain attenuation; Cataldo & Harris, 1982), while others have suggested that this class of SIB could be an elicited response (i.e., an unconditioned frustrative response, or established via Pavlovian conditioning; Furniss & Biswas, 2012). Automatically reinforced SIB (ASIB) has long been considered the least understood and most challenging type of SIB, largely because the events that occasion and maintain it are not known.

Whereas ASIB is often assumed to be under greater relative control by biological variables, socially-maintained SIB is thought to be under greater control by environmental variables. Recent research delineating subtypes of ASIB, including one that is amenable to change and one that is highly invariant, further subcategorizes ASIB and enables classification of SIB across a broader continuum of relative environmental-biological influence (Hagopian, Rooker, Zarcone, 2015; Hagopian, Rooker, Zarcone, Bonner, & Arevalo, 2017). ASIB subtypes are defined based on criteria quantifying the relative difference in the rate of SIB across high and low stimulation conditions of the functional analysis. Subtypes cannot be derived using indirect functional assessment rating scales or questionnaires because these methods do not provide direct observation data on the rate of SIB across these conditions. Subtype-1 ASIB is characterized by higher rates of SIB in conditions with minimal external stimulation, and by lower rates in the play condition (an enriched environment). Subtype-2 ASIB is characterized by high and sometimes variable rates of SIB across high and low stimulation conditions. Thus, Subtype-1 ASIB is sensitive to disruption by alternative reinforcement whereas Subtype-2 is generally invariant. Subtype-3 ASIB is characterized by the presence of self-restraint, a behaviour that is incompatible with SIB and includes wrapping body parts using clothing, continuously holding objects, and using others to restrict movements (Oliver, Murphy, Hall, Arron, & Leggett, 2003). The presence of selfrestraint could suggest that SIB may be reinforced by one mechanism but produces aversive consequences (such as pain) by another, which is avoided through self-restraint (Fisher & Iwata, 1996).

In the first study describing these subtypes of ASIB in hospitalized patients (n = 39 with ASIB; and 13 controls with socially-maintained SIB), subtypes were found to differ markedly in terms of their response to treatment, and across other dimensions (Hagopian et al., 2015). Subtype-1 ASIB and socially-maintained SIB were both found to be highly

responsive to treatment involving reinforcement alone: an 80% reduction in SIB was achieved in 75% cases with Subtype-1 ASIB, and in 84.6% of cases with sociallymaintained SIB. In contrast, Subtype-2 ASIB was found to be highly resistant to treatment involving reinforcement alone: an 80% reduction in SIB was not achieved with any cases. For those with Subtype-2, blocking, punishment, restraint, protective equipment, or some combination was necessary (in addition to reinforcement). Reinforcement alone was not even attempted for Subtype-3 because it was unsafe to evaluate it without the use of restraint or protective equipment. Those with Subtype-2 emitted SIB at significantly higher rates during the play condition which, again, represents an enriched environment (M = 8.6 responses per min; rpm), relative to those with Subtype-1 (M = 1.8 rpm) or socially-maintained SIB (M = 0.3 rpm). Moreover, most individuals with Subtype-1 and socially-maintained SIB (R7.5% and 92.3\%, respectively) displayed other forms of problem behaviour, such as aggression and disruptive behaviour (which were more likely to be maintained by social reinforcement), whereas only roughly half of those with Subtype-2 or -3 ASIB engaged in problem behaviour other than SIB.

A subsequent study examining every published dataset of ASIB with sufficient data to permit subtyping (n = 49 with ASIB; and 13 controls with socially-maintained SIB) largely replicated the findings of the first study with respect to Subtypes-1 and -2 (Hagopian et al., 2017). This sample was comprised of a diverse group, including participants treated in five types of settings (e.g., outpatient clinics, residential programs), described in studies published over a 20 year span, and authored by researchers affiliated with 15 different institutions. Although self-restraint (a defining feature of Subtype-3) was reported in many studies, only a few included sufficient data to enable formal subtyping of these cases. Additional research is needed to confirm whether Subtype-3 ASIB as a distinct subtype, and to better understand self-restraint and its relation to SIB. As was found in the original study, Subtype-2 ASIB was highly resistant to treatment relative to Subtype-1 ASIB and sociallymaintained SIB. Reinforcement alone was effective in 94.1% and 100% of cases with Subtype-1 ASIB and with socially-maintained SIB, respectively, but in only 7.7% of cases with Subtype-2 ASIB. The relative prevalence of Subtypes-1, -2, and -3 ASIB was 41%, 38.5%, and 20.5% (respectively) in the first study where participants were drawn from an hospital sample (Hagopian et al., 2015); and 51%, 44.9%, and 4.1% (respectively) in the replication study (Hagopian et al., 2017). To date, no cases with Subtype-1 ASIB have been admitted to the hospital program that was the site of the original study since 2013, whereas those cases Subtype-2 ASIB (and Subtype-3) are frequently admitted, perhaps suggesting that those with Subtype-1 are being successfully treated as outpatients.

The findings of the replication study (Hagopian et al., 2017) provide support for the generality of the subtyping model. Participants clustered into distinct groups with regard to the level of differentiation of SIB in the functional analysis in a highly comparable way across both studies. Collectively, the findings from both studies (total n = 105 cases where treatment data were available) showed dramatic differences in the responsiveness of these classes of SIB to treatment: reinforcement alone was effective in 79.3% of cases with Subtype-1 ASIB and 93.1% of cases with socially-maintained SIB, but in only 5.0% of cases with Subtype-2 ASIB. A more recent study combining data from both studies by Hagopian and colleagues (Hagopian et al., 2015; 2017) showed classification as Subtype-1 to be a

predictive behavioral marker for response to treatment using reinforcement alone (positive predictive value of 82.6%, negative predictive value of 92.6%; Hagopian, Rooker, & Yenokyan, in press)

#### Implications for Research

The recent identification of Subtype-1 and -2 ASIB, when viewed along with known classes of socially-maintained SIB, allows classification of SIB across a continuum of relative environmental-biological influence. On one end of the continuum where environmental variables have a relatively strong influence, are the socially-maintained functional classes of SIB (where attention, escape, and access to preferred items reinforce SIB). Sociallymaintained functional classes of SIB are highly amenable to behavioural treatment (Greer, Fisher, Saini, Owen, & Jones, 2016). Subtype-1 ASIB is further along the continuum toward greater relative influence by biological variables. Although Subtype-1 occurs independent of social reinforcement, it changes across conditions of high and low external stimulation. The manner in which Subtype-1 varies inversely with the availability of alternative reinforcement is a well-established characteristic of a reinforced response, perhaps suggesting Subtype-1 produces sensory reinforcement. Treatment involving reinforcement may simply provide alternative sources of sensory stimulation that successfully compete with the stimulation produced by SIB. Shifting still further on the continuum is Subtype-2 ASIB where the relative influence of biological variables appears predominant and influence by environmental variables is diminished. The insensitivity of Subtype-2 ASIB to changes in the environment (in both the assessment and treatment contexts) suggests alternative sources of reinforcement cannot compete with whatever occasions and/or maintains Subtype-2 ASIB. It is possible that Subtype-2 ASIB: a) may produce highly potent biologically reinforcing consequences (e.g., endogenous opioids; Cataldo & Harris, 1982); b) may be related to aberrant sensory function, including nocicepetion (Symons, 2011); or c) could be related to some type of motoric dysfunction yet to be determined resulting in repetitive behaviour, perhaps related to Tourette syndrome or obsessive compulsive disorder (Muehlmann & Lewis, 2012). What is relatively clear about Subtype-2 ASIB is that its general insensitivity to changes in the environment makes it a unique among other classes of SIB (and problem behaviour in general), suggesting it is driven primarily, if not exclusively, by biological variables.

Although complex interactions and developmental processes that lead to the emergence of SIB are not fully understood, there is sufficient empirical support and the methodology exists to classify SIB across a broad continuum of relative environmental-biological influence. Socially-maintained SIB, Subtype-1 ASIB, and Subtype-2 ASIB appear to be highly distinct functional classes of SIB that differ on many dimensions. They differ in terms of their sensitivity to change related to social variables and environmental stimulation, and with respect their response to treatment, their frequency, and co-occurrence with other behaviours. Differences between these functional classes are robust and clinically relevant. Subtype-2 ASIB, the most invariant and treatment-resistant class of SIB, appears to represent a unique functional class that could be primarily under control of biological variables. Having identified Subtype-2 ASIB, the need to understand the biological

Hagopian and Frank-Crawford

mechanisms underlying this subtype and develop interventions for it is now a clinical and scientific imperative.

Although behaviour analytic research typically identifies the function of SIB and uses that to inform treatment selection, additional research such as those discussed here (e.g., Hagopian et al., 2015; 2017) are needed to examine how SIB differs across functional classes, including whether certain types of treatments are more effective for one class relative to another. Research on the biology, phenomenology, and pharmacological treatment of SIB would be greatly enhanced if it controlled for and directly examined functional classes of SIB. Most studies on these topics do not report on the functional class of SIB, so their participants likely include individuals with socially-maintained SIB and ASIB of all subtypes. A recent review of research on naltrexone (Roy, Roy, Deb, Unwin, & Roy, 2015) examined outcomes reported in 10 randomized clinical trials. The authors found that only 50% of individuals showed a reduction in SIB and speculated that some of the nonresponders could have been individuals whose SIB was maintained by environmental variables. These comments echo those of Symons, Thompson, and Rodriguez (2004) who conducted a 20-year review of 27 naltrexone studies 10 years prior and concluded that classifying SIB based on its function could reduce heterogeneity in pharmacological research. Unfortunately, post-hoc analysis of the role of functional class of SIB in past naltrexone trials is not possible because the majority of these studies do not report on the functional classification of SIB (though exceptions exist, Garcia & Smith, 1999). Likewise, the vast majority of trials examining the effects of other classes of medications on SIB do not report on its function class and likely include similarly heterogeneous samples (e.g., Ruedrich et al., 2008). Future pharmacological studies could, at minimum, identify the functional class of SIB and control for this variable. A better alternative would be select certain functional classes of SIB for comparison and examine differential responsiveness to medication. It is possible that certain medications are differentially effective for certain functional classes. However without controlling for this variable, those effects are diminished or perhaps obscured completely due to participant heterogeneity. Studies examining the phenomenology of SIB, risk factors, and role of pain and other biological variables in SIB could also control for and examine the effects of the functional class of SIB. Given how distinct these classes of SIB appear to be in terms of their clinical presentation, such an analysis has the potential to identify important differences.

Studies need not examine or compare all functional classes of SIB; rather, comparisons of at least two classes might be sufficient depending upon the question. For example, comparison of participants with socially-maintained SIB versus those with Subtype-2 ASIB might be useful as these two functional classes represent extreme ends of the continuum of relative environmental-biological influence. Comparisons across Subtype-1 and -2 ASIB might be more useful to examine the mechanisms underlying Subtype-2 ASIB – a particularly important endeavor because this functional class of SIB currently represents the most treatment-resistant and severe class of SIB that is known. Analysis of SIB as distinct functional classes would not only control for sources of variance, but would also align research with conceptual models that view SIB as the product of interactions between biological and environmental variables. Adopting this approach also has the potential to

Hagopian and Frank-Crawford

The value of classifying problems based on an understanding of their underlying mechanisms is important to structuring research and advancing knowledge. This is perhaps best illustrated with the example of precision medicine, made possible with advances in genomics and other technologies that enable researchers to examine diseases at the molecular level (National Research Council, 2011). This approach recognizes that seemingly similar diseases can have subtypes when examined at a more molecular level; and has recently been applied to SIB (Hagopian, et al., in press). The traditional model of disease classification in terms of signs and symptoms is giving way to a model based on the causal mechanisms of diseases, and identification of disease subtypes, thus informing individualized care based on subtype and other individual differences. Although the analogy of precision medicine is not perfectly applicable to SIB, as the causal mechanisms of SIB cannot be reduced to genomic variants, it alludes to the potential benefits of conceptualizing and classifying SIB based on an analysis of its current controlling variables. If we are to advance knowledge of SIB and develop new and more efficacious treatments for it, we must recognize that SIB is not a single entity, but comprised of distinct classes.

#### Acknowledgments

Manuscript preparation was supported by Grant R01 HD076653 from the National Institute of Child Health and Human Development (NICHD) and U54 HD079123 from the Intellectual and Developmental Disabilities Research Centers (IDDRC). The contents are solely the responsibility of the authors and do not necessarily represent the official views of NICHD or IDDRCs.

The authors would like to thank Meagan K. Gregory and Griffin W. Rooker for their assistance with this manuscript.

## References

- Beavers GA, Iwata BA, Lerman DC. Thirty years of research on the functional analysis of problem behavior. Journal of Applied Behavior Analysis. 2013; 46:1–21. DOI: 10.1002/jaba.30 [PubMed: 24114081]
- Cataldo MF, Harris J. The biological basis for self-injury in the mentally retarded. Analysis and Intervention in Developmental Disabilities. 1982; 2:21–39. DOI: 10.1016/0270-4684(82)90004-0
- Fisher WW, Iwata BA. On the function of self-restraint and its relationship to self-injury. Journal of Applied Behavior Analysis. 1996; 29:93–98. DOI: 10.1901/jaba.1996.29-93 [PubMed: 8881347]
- Furniss F, Biswas AB. Recent research on aetiology, development and phenomenology of selfinjurious behaviour in people with intellectual disabilities: a systematic review and implications for treatment. Journal of Intellectual Disability Research. 2012; 56:453–475. DOI: 10.1111/j. 1365-2788.2012.01534.x [PubMed: 22369696]
- Garcia D, Smith RG. Using analog baselines to assess the effects of naltrexone on self-injurious behavior. Research in Developmental Disabilities. 1999; 20:1–21. DOI: 10.1016/S0891-4222(98)00028-6 [PubMed: 9987807]
- Greer BD, Fisher WW, Saini V, Owen TM, Jones JK. Functional communication training during reinforcement schedule thinning: An analysis of 25 applications. Journal of Applied Behavior Analysis. 2016; 49:105–121. [PubMed: 26482103]
- Hagopian LP, Rooker GW, Yenokyan G. Identifying predictive behavioral markers: A demonstration using automatically reinforced self-injurious behavior. Journal of Applied Behavior Analysis. (in press).

Hagopian and Frank-Crawford

- Hagopian LP, Rooker GW, Zarcone JR. Delineating subtypes of self-injurious behavior maintained by automatic reinforcement. Journal of Applied Behavior Analysis. 2015; 48:523–543. DOI: 10.1002/jaba.236 [PubMed: 26223959]
- Hagopian LP, Rooker GW, Zarcone JR, Bonner AC, Arevalo AR. Further analysis of subtypes of automatically reinforced SIB: A replication and quantitative analysis of published datasets. Journal of Applied Behavior Analysis. 2017; 50:48–66. DOI: 10.1002/jaba.368 [PubMed: 28032344]
- LeBlanc LA, Patel MR, Carr JE. Recent advances in the assessment of aberrant behavior maintained by automatic reinforcement in individuals with developmental disabilities. Journal of Behavior Therapy and Experimental Psychiatry. 2000; 31:137–154. DOI: 10.1016/S0005-7916(00)00017-3 [PubMed: 11132117]
- Muehlmann AM, Lewis MH. Abnormal repetitive behaviours: Shared phenomenology and pathophysiology. Journal of Intellectual Disability Research. 2012; 56:427–440. DOI: 10.1111/j. 1365-2788.2011.01519.x [PubMed: 22283923]
- National Research Council. Toward precision medicine: Building a knowledge network for biomedical research and a new taxonomy of disease. Washington DC: National Academies Press; 2011.
- Oliver C, Adams D, Allen D, Bull L, Heald M, Moss J, Woodcock K. Causal models of clinically significant behaviors in Angelman, Cornelia de Lange, Prader-Willi and Smith-Magenis syndromes. International Review of Research in Developmental Disabilities. 2013; 44:167–212. DOI: 10.1016/B978-0-12-401662-0.00006-3
- Oliver C, Murphy G, Hall S, Arron K, Leggett J. Phenomenology of self-restraint. American Journal on Mental Retardation. 2003; 108:71–81. DOI: 10.1352/0895-8017(2003)108<0071:POSR>2.0.CO;2 [PubMed: 12564940]
- Richman DM, Lindauer SE. Longitudinal assessment of stereotypic, proto-injurious, and self-injurious behavior exhibited by young children with developmental delays. American Journal on Mental Retardation. 2006; 111:137–137. DOI: 10.1352/0895-8017(2005)110[439:LAOSPA]2.0.CO;2
- Roy A, Roy M, Deb S, Unwin G. Are opioid antagonists effective in reducing self-injury in adults with intellectual disability? A systematic review. Journal of Intellectual Disability Research. 2015; 59:55–67. DOI: 10.1111/jir.12111 [PubMed: 24397316]
- Ruedrich SL, Swales TP, Rossvanes C, Diana L, Arkadiev V, Lim K. Atypical antipsychotic medication improves aggression, but not self-injurious behaviour, in adults with intellectual disabilities. Journal of Intellectual Disability Research. 2008; 52:132–140. 10.1111.j. 1365-2007.00981.x. [PubMed: 18197952]
- Symons FJ. Self-injurious behavior in neurodevelopmental disorders: Relevance of nociceptive and immune mechanisms. Neuroscience & Biobehavioral Reviews. 2011; 35:1266–1274. DOI: 10.1016/j.neubiorev.2011.01.002 [PubMed: 21237197]
- Symons FJ, Thompson A, Rodriguez MC. Self-injurious behavior and the efficacy of naltrexone treatment: A quantitative synthesis. Mental Retardation and Developmental Disabilities Research Reviews. 2004; 10:193–200. DOI: 10.1002/mrdd.20031 [PubMed: 15611982]