**Article title:** ASD intervention research in real world contexts: Refining single case designs

**Journal title:** Research in Autism Spectrum Disorders

**Corresponding author:** Ms. Kim Bulkeley

**Final version published online:** 13-AUG-2013

**Full bibliographic details:** Research in Autism Spectrum Disorders 7 (2013), pp. 1257-1264

**DOI information:** 10.1016/j.rasd.2013.07.014

**Published version of the article is available on:**

ASD intervention research in real world contexts: Refining single case designs

Kim Bulkeley¹
Anita Bundy¹
Jacqueline Roberts²
Stewart Einfeld¹

¹Faculty of Health Sciences, University of Sydney; ²Griffith University

Keywords: autism; social validity; repeated measures; individualized intervention; outcome measurement; intervention fidelity

Abstract:

There is a pressing need for intervention research that reflects real world practice to support evidence based decision making for families, professionals and administrators who support children with ASD. Some of the challenges confronting intervention research are explored, with solutions offered based on single case design methodology. Challenges with single case designs are also outlined and contemporary solutions that are applicable in real world contexts are illustrated in a study by the authors. Research utilizing these strategies may assist with facilitating practitioners to engage in practice-based research to bridge the research to practice gap in intervention with young children with ASD.
Intervention research that reflects real world contexts is required by service users, professionals, and policy makers to understand how to promote the best outcomes for individuals with autism spectrum disorders (ASD) and to decide which interventions will be funded using public money. The presentation of children with ASD has grown significantly (Matson & Kozlowski, 2011), with a prevalence of 1 in 88 recently reported in the United States (Centers for Disease Control and Prevention, 2012). The cost to governments and communities who support those with ASD is significant with individual estimates as high as $3.2 million throughout the lifespan (Ganz, 2007). Effective interventions, particularly if provided early, can improve outcomes, reducing the cost of support by up to 65% across the life-course (National Autism Centre, 2009).

A plethora of reviews of intervention evidence (Klintwall, Gillberg, Bolte, & Fernell, 2012; LeBlanc & Gillis, 2012; National Autism Centre, 2009; Prior, Roberts, Rodger, Williams, & Sutherland, 2011; Roberts & Prior, 2006) describe interventions that meet some of the needs of some children with ASD (Stahmer, Schreibman, & Cunningham, 2011). However, we lack information to support many of the interventions in current use and recent findings suggest that families and professionals regularly use a wide range of interventions with little or no empirical support (Carter, et al., 2011; Goin-Kochel, Myers, & Mackintosh, 2007; Hess, Morrier, Heflin, & Ivey, 2008; Stahmer, 2007). Clearly, more research into contemporary interventions is required.

Intervention research for all children with disabilities is notoriously difficult, lags behind the experience of families and professionals, and is not readily translated into real world usage (Odom, Hume, Boyd, & Stabel, 2012). In a review of scientific methods in special education, including children with autism, Odom et al (2005) claimed that research in this field is “hardest-to-do science” reflecting the complexity of the participant group.
Randomized control trials (RCTs) are regarded by some as gold standard evidence, making an important contribution to evidence-based practice. A basic premise of RCTs is the random allocation of participants to one of two groups, one that receives the intervention under scrutiny and the other that does not. Pre- and post-testing is conducted and comparisons of the group data occurs to determine if greater gains were obtained for the treatment group over the control group. However, RCTs are expensive and require large numbers of participants. Further, many interventions used with children with ASD do not lend themselves readily to RCTs. Nonetheless, the high regard of RCTs has led to an undervaluing of other research designs and, consequently, fewer studies of the wide variety of interventions in current use in the ASD field (Smith, et al., 2007).

Regardless of design, the resources required to conduct research interfere with the comprehensive investigation of interventions for children with ASD (Mesibov & Shea, 2011). Time and money are spread thinly across a diverse range of areas, highlighting the need for research designs that use minimal additional resources, produce robust results and yield practice-ready outcomes. Research conducted in real world settings has the potential to spread limited resources further and improve the usability and quantity of intervention research (Jenkins, Price, & Straker, 1998). Embedding intervention research into practice settings has the secondary benefit of minimising the resources required from both participants and researchers.

In addition to the struggle to find resources, the ecological and social validity of research evidence is another important consideration for service providers who support the health, community inclusion and education of young children with ASD (Dingfelder & Mandell, 2011).
Testing of interventions in controlled experimental environments prior to application in real world settings (i.e., the efficacy-to-effectiveness pipeline) is viewed widely as rigorous research process (Smith, et al., 2007). However, the attempt to follow rigorous practices may lead researchers to extract narrow versions of interventions used in practice and therefore divorce the intervention from a real-world context. The identification of research designs that are scientifically sound as well as ecologically and socially valid is a challenge facing contemporary intervention researchers (Dingfelder & Mandell, 2011).

A research frame is needed that allows eclectic approaches that are individually tailored; includes socially valid outcome measures (Mesibov & Shea, 2011); and is not resource intensive. Single case design (SCD) research is often touted as a way to solve some of the problems with ASD intervention research and, indeed, it can do that. SCD takes the individual as the focus, measuring target behaviors repeatedly across phases as the independent variable is manipulated. This establishes individual participants as their own controls and allows functional relationships to be observed between behavior change and intervention (Kazdin, 2011). The focus on the individual is an advantage in ASD research due to resource issues, the heterogeneity of the population and the need to individualize interventions (Mesibov & Shea, 2011). However SCDs are also associated with particular research challenges when applied to young children with ASD.

While the problems encountered in research evaluating interventions for children with ASD are common across all research designs and no one design can claim exclusive superiority, SCDs show promise for contributing to the evidence base. SCDs have been proposed as a robust
mechanism for intervention research in a range of professions including occupational therapy (Johnston & Smith, 2010), special education (R. Horner, et al., 2005; Kratochwill, et al., 2010) and psychology (Levin, Ferron, & Kratochwill, 2012). SCDs include planned management of the independent variable while repeatedly monitoring the dependent variable, thereby differentiating SCDs from case studies which are a descriptive report on the experience and outcomes of individuals. Originating in the applied behaviour analysis field, SCDs are now widely used; have been included in recent ASD intervention research reviews (National Autism Centre, 2009); feature in discussion on evidence based practice in ASD (Matson, Turygin, Beighley, & Matson, 2012) and are gaining support as a source of high level evidence in intervention research (Kratochwill, et al., 2010; Perdices & Tate, 2009). Awareness of the value of this methodology has led to the development of tools for scrutinising SCD quality (Kratochwill, et al., 2010; Logan, Hickman, Harris, & Heriza, 2008; Tate, et al., 2008) and ongoing efforts to develop systems for aggregating the findings of this growing body of research through meta-analysis (Parker, Vannest, & Davis, 2011; Wolery, Busick, Reichow, & Barton, 2010). A comprehensive treatment of SCD approaches can be found in texts by Kazdin (2011) and Gast (2010) devoted to this topic specifically.

In this paper, we focus on: (a) issues readily addressed by SCD research and (b) problems associated with SCD research. We conclude with an example of an SCD illustrating solutions to common problems.

**Issues readily addressed by SCD research**

SCDs can be undertaken in real world settings, increasing the social validity of the findings. They eliminate the need for control groups and require fewer participants than RCTs
and other experimental designs. They allow for the individualization of interventions and, consequently contribute to a greater understanding of individual responses to an intervention.

*Real world practice and social validity*

SCD research is a method that can be applied in practice and community settings with minimal alteration to the way intervention is provided. The nature of SCD, with a focus on individual participants, allows for the establishment of outcome measures and targets of intervention that are aligned with the preferences and priorities of participants, families and other service providers, thus enhancing the social validity of the research (Perdices & Tate, 2009).

*Elimination of control groups*

Because individual participants engage in multiple SCD phases, they serve as their own controls. Thus, the ethical issues of withholding treatment are minimized or removed as all participants receive treatment in the course of the study with relatively brief delays in access (Lord, et al., 2005). Furthermore, if there is risk of harm from delaying treatment, as in the case of self injurious behavior, there are SCD options that remove an initial pre-intervention baseline period, providing comparison treatment phases of a brief duration.

*Small Numbers of Participants*

SCDs can yield robust findings with a small number of participants, solving problems with recruitment that abound in ASD research with children. Due to the heterogeneous nature of ASD, recruitment of large numbers of participants that are sufficiently similar can be problematic. SCDs require a small number of participants, making it easier to focus on a clearly defined participant group and to apply narrow inclusion criteria with some confidence of recruiting adequate numbers of participants to make the design work. Further, families with
children with ASD have significant levels of stress (Lovell, Moss, & Wetherell, 2012) and are reported to engage in multiple and intensive interventions (Green, et al., 2006). Research activity, no matter how important, adds to their responsibilities, reducing the likelihood of participation and highlighting the need for designs like SCD that require fewer participants to yield robust outcomes. SCD research is easily embedded into practice, making it possible to generate research evidence while providing services that participants engage in as part of their regular routine with minimal additional demands on participants. Recruitment to these designs is likely to be less problematic when they are embedded in real world practice.

While SCDs are sometimes criticized because small numbers can limit generalizability (Smith, et al., 2007), external validity can be strengthened through some relatively simple procedures: including multiple participants (at least 3 replications across participants), clearly describing participants, operationalizing the context where intervention is provided, and replicating the study across sites with independent research groups (R. Horner, et al., 2005). As external validity improves so also does the quality of the evidence base for ASD interventions using SCDs.

**Individualizing of Interventions**

SCDs allow for the development of individualized protocols (independent variable) within a consistent framework for comparison across participants (Mesibov & Shea, 2011). Individualized variation in intervention for young children with ASD is regarded as best practice (Barton, Lawrence, & Deurloo, 2012; Delmolino & Harris, 2012; Strain, Schwartz, & Barton, 2011); a one-size-fits-all approach is rarely appropriate. In SCD, a hypothesis is formed from careful observation about the best strategies to apply within the overall intervention framework.
Ongoing monitoring to track individual responses and outcomes is inherent to SCD. Articulating consistent intervention principles as part of the protocol allows for replication of the study (Mesibov & Shea, 2011).

*Understanding individual treatment response*

The nature of ASD as a spectrum disorder implies heterogeneity of individuals with the diagnosis and forewarns of the need to understand individual responses to intervention. The individual treatment (dependent variable) response story is not well told by group designs where participant data are aggregated to determine effectiveness, masking much of the variability that is often present (R. Horner, et al., 2005; Smith, et al., 2007). SCDs focus on the individual as the unit of concern, and report specific and detailed information on the response of individual participants on repeated occasions throughout the phases of the research. This information may establish a functional link between treatment and response, generate hypotheses about differential treatment responses, inform intervention decisions, provide empirical support for current practice and highlight targets for future research (Kazdin, 2011).

*Problems associated with SCD research*

SCDs, like other research designs, are not without limitations. The methodology is emerging and is not strongly embedded in undergraduate training or the broader discourse of intervention research. This can lead to reluctance to conduct SCDs and the need to encourage both participants and practitioners/researchers to understand and accept the contributions of this methodology (Perdices & Tate, 2009). Further, the same problems common to the conduct of all ASD intervention research also apply to SCDs: having tools appropriate for repeated
measurement, rater drift, rater bias, complications of multiple raters (reliability), contextualising non-standardised ratings, the capacity to withdraw an intervention, measurement of intervention fidelity and assessment of maintenance of gains.

Repeated measurement tools

A key feature of SCD research is the need for short-interval, repeated measurement of the dependent variable. The repeated measure must be user-friendly, efficient, consistent, and reduce the imposition of frequent ratings, especially in natural contexts. Many mechanisms have been used previously: manually recorded behavioral observations (Dykstra, Boyd, Watson, Crais, & Baranek, 2012), computer-based recording of observations (Gast, 2010), video recordings (Boyd, McDonough, Rupp, Khan, & Bodfish, 2011), and partial interval recording (Davis, Durand, & Chan, 2011). These all require significant resources, both at the point where intervention is provided and in analysis of the data. In real world contexts, where practitioner researchers work in home or community settings, research is not the primary focus and it is not possible to use measurement tools that require this level of resourcing.

The engagement of parents or carers in the repeated measurement of behavior and the development of outcome measures that are sensitive and specific to the needs of particular children is emerging as an important part of designing intervention research (Adams, et al., 2012). SCDs in ASD may benefit from measurement approaches that embrace this perspective, generating evidence that is defensible and readily applied in real world settings.

Rater Drift

Rater drift is another important issue that requires careful consideration in repeated measurement (Wewers & Lowe, 1990). The tendency to change the focus from the original target of concern as priorities change presents a particular challenge in SCDs; measurement is
frequent and the overall behavior of the participant is subject to intense scrutiny, both of which may contribute to an increased tendency for the rater to drift from the original behavior of interest. Measurement tools that incorporate a clear reminder of the target behavior are essential to ensure consistency in the recognition of the behavior of concern (Gast, 2010).

**Rater bias**

Rater bias, also known in this case as a placebo effect, has been recognized as an important consideration in intervention research, particularly when using parent-report outcome measures (Lord, et al., 2005). For any number of reasons, parents of young children with ASD are likely to report positive outcomes for the majority of interventions (Goin-Kochel, Mackintosh, & Myers, 2009). Many SCDs offer a single intervention sandwiched between baseline phases. Blinding of raters, essential to the objectivity of repeated ratings, is not generally possible in SCDs. Rater bias is potentially heightened when parents and carers rate a child’s performance in natural contexts.

**Multiple raters**

The problems associated with multiple raters are heightened in SCDs due to the frequency of behavior rating. This is generally managed by training raters and comparing ratings until acceptable agreement is obtained between raters (Gast, 2010; Ottenbacher, 1986). In real world settings, however, engaging multiple raters and repeated training imposes a significant burden. One option, commonly employed in SCD, is to remove the rating of performance from the practice setting by training raters who are independent to the provision of the intervention to rate from videotape (Kazdin, 2011). However, this may not be a viable solution when project resources cannot sustain the imposition of repeated video recording and the lengthy process of rating video excerpts.
The engagement of parents and carers as raters provides an important source of information about the lived experience of the performance of the child (Adams, et al., 2012) and the social and ecological validity of the changes in behavior as a result of interventions (Chafouleas, Sanetti, Kilgus, & Maggin, 2012). However, there are limitations to data gained in this way as multiple informants have been reported to vary in their ratings of an individual child’s behavior (Achenbach, 2011; Achenbach, Edelbrock, & Howell, 1987). SCD studies must explore options for reducing or managing the effects of multiple raters on repeated measure data.

Contextualizing of observations

Individualized and repeated observation is a feature of SCDs that is highly valued for its social and ecological validity (Perdices & Tate, 2009). Standardized instruments may not be applicable in SCDs due to limitations in their re-application over short intervals and a lack of relevance of items. However, the interpretation of outcomes can be greatly enhanced by understanding how the behaviors represented by the observations compare to similar behaviors of a larger group; such contextualizing may provide insight into the significance of changes in behavior over time (Johnston & Smith, 2010). Standardized behaviour rating scales can be incorporated into an SCD at baseline and follow up to contextualize and supplement the individualized repeated observations.

Inability to Withdraw Intervention

Demonstration of a functional link between behavior change and the introduction of an intervention is the essence of intervention research. Because there is no understanding of the impact of removal of the independent variable in SCDs where a baseline phase is followed only by a single intervention phase (i.e., A-B), the functional linkage is relatively weak (Johnston &
Smith, 2010). When a return-to-baseline phase is included (i.e., A-B-A) showing behavior returning to pretreatment levels (Kazdin, 2011), the functional link between behavior change and intervention is strengthened. However, when interventions make permanent changes (e.g., building skills), often the case with interventions delivered to children with ASD (Lord, et al., 2005), the removal of intervention cannot demonstrate a functional link. Thus, there is a need to find other ways of demonstrating a connection between intervention and the behavior change.

Multiple baseline designs, where there are multiple participants or where an intervention is introduced across several behaviors with one participant provide a means of strengthening the linkage between intervention and behavior change (Kazdin, 2011). Multiple baselines across behaviors may be problematic, however, when the interventions involve building the skills of parents or carers, who may inadvertently apply all their newfound skills in all situations. Exploring combinations of multiple baseline and alternating treatment designs (Kazdin, 2011) may provide an option to counteract this difficulty. (Leemrijse, Meijer, Vermeer, Ader, & Diemel, 2000).

*Intervention Fidelity*

Measurement of intervention fidelity, commonly overlooked in research, confirms that participants receive an intervention as proposed (Wolery, 2011). In addition, measurement of fidelity ensures transparency in the differentiation of intervention types when two or more interventions are applied (Kazdin, 2011), thus strengthening claims of influence on outcomes (S. Horner, Rew, & Torres, 2006). Individualized interventions also challenge the measurement of fidelity and heighten the need for clear processes for confirming the nature of interventions provided in SCD research protocols.
Maintenance of change

Understanding the persistence of behavior change following intervention is a crucial aspect of intervention research. SCD research generally focuses on short term interventions and immediate measures of outcome which provide little information about the maintenance over time of observed changes in behavior (Smith, et al., 2007). Intensive observation of behaviors inherent to SCDs, are difficult to sustain. Nonetheless, SCD research requires follow up that does not create undue demands on participants or practitioners.

A contemporary SCD

A contemporary SCD from a study by the authors, illustrates solutions to some of the SCD research challenges outlined in this paper (Table 1). This design is discussed below, presenting: a repeated measurement option; the use of standardised testing; the application of two interventions to strengthen the design; the inclusion of a probe phase; and a feasible method for measurement of intervention fidelity.
Measurement issues

The identification of a suitable repeated measurement tool was resolved by modifying a visual analogue scale (VAS) which has been used successfully in a variety of acute and community care settings (Wewers & Lowe, 1990) and, more recently, in SCDs (Hoogeboom, et
al., 2012). VAS is inexpensive, individualized, flexible, defensible, efficient and user friendly. VASs are also readily transferred onto a variety of electronic and hard copy platforms for simple and accurate completion (Dale & Hagen, 2007; Wewers & Lowe, 1990).

A VAS was developed as the repeated measurement tool for this study to be completed by the same parent on a daily basis throughout the phases of the protocol. The VAS was used to identify their perception of the level of the target behaviour on each day. The target behavior was selected by the parent and individualized for each participant. A clear description of the behavior was provided on each VAS, managing rater drift. A pilot study conducted by the authors indicated feasibility of VAS from the family perspective but also highlighted some practical issues with the completion of the scales in a paper format. These scales are easily generated using electronic platforms, which promote accuracy of completion and the transfer of responses to a digital data format. The Experience Sampling Program\textsuperscript{TM} (ESP\textsuperscript{TM}) was selected as a platform for individually programming personal digital assistants which were inexpensive and readily available at the time of the study. (Note, this type of programming is also available for smartphones and touch screen devices). Rater discrepancies were avoided by engaging a single rater (Chafouleas, et al., 2010) for the duration of the study.

\textit{Standardized pre- and post- measurement}

To supplement and contextualize individualized parent ratings of the target behaviour, parents completed the Developmental Behaviour Checklist-Parent version (DBC-P) (Einfeld & Tonge, 2002). Parents completed the rating prior to the first baseline and at follow up.

\textit{Conducting two interventions from different theoretical perspectives}

By including two potentially effective interventions (a combination of multiple baseline design and alternating treatment design) applied in a random order, we managed threats to
experimental control and treatment order interference. The protocol would be described as ABAC or ACAB in SCD terminology, where A is baseline, B is intervention type 1 and C is intervention type 2. This design also promoted rater blinding as the participants and their carers were not sensitized to a preferred intervention. This design accommodated the inability to withdraw the intervention and allowed for stronger comparison of differential intervention effects within an SCD framework.

*Probe phase*

The maintenance of improvements over time is highly valued and an important consideration in the evaluation of evidence in support of intervention effectiveness. Historically SCDs offered only a short term view of behavior change; however more contemporary designs are examining maintenance of change. The inclusion of a probe phase (Finnigan & Starr, 2010; Gutman, et al., 2010), in which we resumed the repeated observation after a break, provided an option for long-term follow up of the target behavior. A probe phase maintains the integrity of the repeated measurement, by extending the period over which outcomes are measured. The resultant protocol adopted for our intervention study became ABAC-A or ACAB-A with randomisation to order of interventions, with the final baseline (A) phase occurring 6 months after the second intervention was completed (Table 1).

*Fidelity Measurement*

Treatment fidelity, essential for intervention research, has been underdeveloped in SCD studies. We opted to collect audio recordings of all interventions sessions, which shows promise for intervention fidelity measurement (Bellg, et al., 2004; Torrey, 2012). Audio recordings of intervention sessions require minimal additional resources, are unobtrusively obtained using digital recording devices and provide a good source of material for analysis by independent
raters. Checklists of expected features of the interventions were established for rating of quality and content of interventions by an independent rater and for differentiation of interventions over the course of the phases in our SCD protocol.

**Summary and Conclusions**

We have described (a) common problems that impede the conduct of intervention research for young children with ASD in natural settings, (b) strengths and weaknesses of SCD research and (c) an application of SCD with children with ASD. SCD research is a robust experimental method, providing potential solutions to problems with recruitment, limited funding for research, ethical issues of withholding intervention, individualizing of interventions and understanding of individual intervention responses. SCDs also support the conduct of research in practice environments and captures the expertise of practitioners in the generation of empirical support for interventions (Perdices & Tate, 2009). The refinement of SCD research has the potential to yield a significant body of evidence that is easily interpreted by service users, service providers and service administrators. These research designs will enhance outcomes for young children with ASD by building practice based evidence to support evidence based practice.

Acknowledgements: I would like to thank Elaine Tam for her patience and support with endnote and Marie McInnes for assistance with proof reading.


spectrum disorders. In H. Australian Government Department of Families, Community Services and Indigenous Affairs (Ed.), (pp. 164): Australian Government


