A gradual effects model for single-case designs

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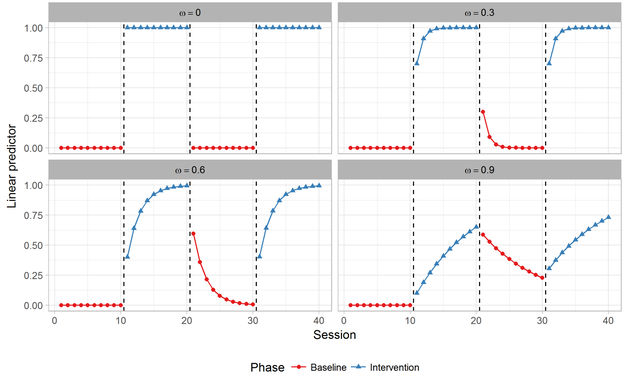
Introduction

Single-case designs play an important role in certain disciplines within education research—particularly as a tool for developing and evaluating treatment practices for individuals with low-incidence disabilities. There has been growing interest in using meta-analysis to synthesize evidence from single-case designs. However, single-case data have a number of features that present challenges for statistical modeling and effect size estimation. Recent critical reviews have identified several criteria that effect size indices should ideally meet in order to be useful for meta-analysis of single-case research:

- account for the presence of time trends (Wolery, Busick, Reichow, & Barton, 2010);
- use all available data, rather than selected subsets of data (Horner, Swaminathan, Sugai, & Smolkowski, 2012; Wolery et al., 2010);
- make appropriate assumptions about distribution of dependent variable (Shadish, 2014);
- account for the possibility of serial dependence among the outcome measurements (Horner et al., 2012; Shadish, 2014; Wolery et al., 2010).

In this work, we address several of these criteria by developing a model for single-case data that exhibit non-linear time trends created by an intervention that produces gradual effects, which build up and dissipate over time. The model expresses a structural relationship between a pattern of treatment assignment and an outcome variable, making it appropriate for both treatment reversal and multiple baseline designs. It is formulated as a generalized linear model so that it can be applied to outcomes measured as frequency counts or proportions, both of which are commonly used in single-case research, while providing readily interpretable effect size estimates such as log response ratios or log odds ratios.

Figure 1. Functional specification of the gradual effects model for differing values of ω, where $\beta_0 = 0$, the equilibrium treatment effect is $\beta_1 = 1$, *and* $m = \infty$. Each plot depicts an ABAB design with ten sessions per phase.



The gradual effects model

The gradual effects model applies to the data series for a single case, which might be one of several within a treatment reversal or multiple baseline design. Notation:

- Y_j denotes the observed outcome at measurement occasion $j = 1,\ldots, J$.
- T_j is an indicator for treatment status at time j, where $T_j = 0$ when treatment does not occur during time j and $T_j = 1$ if treatment occurs during time j.
- $\mu_j = \mathrm{E}(Y_j)$ denotes the mean outcome at time j.
- η_j is the linear predictor at time j, defined by the link function $g(x)$, where $g(\mu_j) = \eta_j$.
- $V(\mu_j)$ is a variance function describing the relationship between the mean and the variance of the outcome.

The gradual effects model is then given by:

$$\eta_j = \beta_0 + \beta_1 \left(\frac{1-\omega}{1-\omega^m}\right) \sum_{i=1}^{j} \omega^{i-j} T_i$$

$$\mathrm{Var}(Y_j) = \sigma^2 V(\mu_j)$$

- $\omega \in [0,1)$ represents the delay of the effect of treatment.
  - When $\omega = 0$, the effect of treatment is immediate.
  - For larger ω, the full effect of treatment is increasingly delayed.
- m is a user-specified parameter describing the number of consecutive treatment sessions at which to estimate an effect size.
- σ^2 represents dispersion of the outcome relative to the Poisson distribution.

Effect sizes

The choice of link function determines the form of the effect size estimated by the gradual effects model.

- With an identity link $g(x) = x$, β_1 is an ***additive effect*** (unstandardized mean difference) of m consecutive treatment sessions.
- With a natural log link $g(x) = \ln(x)$, β_1 is a ***log response ratio*** and $\exp(\beta_1)$ is the multiplicative effect of m consecutive treatment sessions.
- With a logit link $g(x) = \ln(x) - \ln(1 - x)$, β_1 is a ***log odds ratio*** corresponding to m consecutive treatment sessions.

Estimation and software

- We estimate the gradual effects model using maximum quasi-likelihood, with profiling in the non-linear parameter ω.
- Implemented in R package `SingleCaseES` (https://github.com/jepusto/SingleCaseES).
- Also available as an interactive web application (written in Shiny) at https://jepusto.shinyapps.io/gem-scd/.

Example: Thorne (2005)

Thorne and Kamps (2008) used an ABAB design to evaluate the effects of a group contingency intervention on academic engagement and levels of problem behaviors among twelve students at risk for developing behavioral disorders. Frequency of inappropriate behavior was recorded via direct observation of each student during 15-minute academic periods.

We fit the gradual effects model for each of the twelve cases in the study.

- quasi-Poisson variance function because outcome is a frequency count.
- log link, so that the effect size estimates are log-response ratios
- effect sizes estimated for $m = 10$ intervention sessions.
- For comparison purposes, also estimated log response ratios using R_1 estimator (Pustejovsky, 2015), which assumes an immediate change-in-levels.
- Average LRR effect size estimated using a random effects meta-analysis model with robust variance estimation (Sidik & Jonkman, 2006)

Figure 2. Rates of inappropriate behavior from Thorne and Kamps (2008), including fitted values from the gradual effects model.

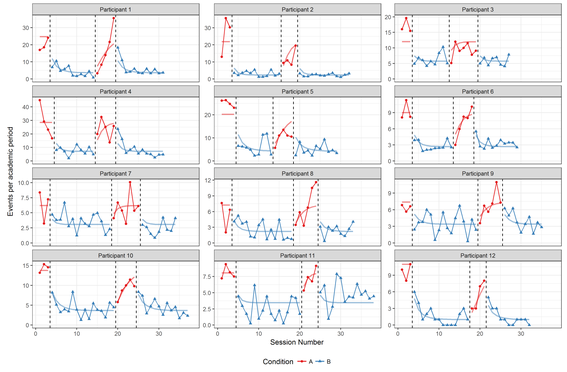


Table 1. Estimates and standard errors for problem behavior from Thorne (2005) from the normally-distributed errors models.

Case	Change-in-levels model LRR est.	SE	Gradual effects model LRR est.	SE	ω	σ^2
Participant 1	-1.22	0.25	-1.91	0.21	0.62	1.47
Participant 2	-1.91	0.24	-2.25	0.22	0.45	1.36
Participant 3	-0.65	0.14	-0.74	0.15	0.35	0.87
Participant 4	-1.17	0.18	-1.38	0.18	0.44	2.18
Participant 5	-1.13	0.20	-1.54	0.25	0.64	1.96
Participant 6	-0.94	0.13	-1.21	0.11	0.49	0.23
Participant 7	-0.63	0.15	-0.70	0.16	0.35	0.66
Participant 8	-0.94	0.21	-1.19	0.24	0.53	0.95
Participant 9	-0.60	.014	-0.77	0.18	0.55	0.70
Participant 10	-0.94	0.13	-1.31	0.13	0.63	0.53
Participant 11	-0.75	0.12	-0.85	0.18	0.35	0.99
Participant 12	-1.50	0.27	-2.38	0.23	0.66	0.66
Random effects meta-analysis	**-0.99**	**0.10**	**-1.34**	**0.16**		

Table 2. Simulation Conditions

Parameter	Levels
Baseline frequency $\exp(\beta_0)$	5, 15, 25
Treatment effect β_1	-1.6 [0.4] 1.6
Delay parameter ω	0.0, 0.3, 0.6, 0.9
Auto-correlation φ	0.0, 0.2, 0.4
Observations per phase	3,5,10

Simulation study

We evaluated parameter recovery in the gradual effects model under conditions similar those seen in applied studies, focusing primarily on accuracy and bias of effect size estimates from GEM. Data were simulated following an ABAB design, using poisson-distributed outcomes and a log-link. We examined both independent outcomes and AR(1) auto-correlated outcomes, generated using binomial thinning (McKenzie, 1988).

- For designs with adequate phase lengths ($n \geq 5$) and baseline frequencies that are not very close to zero, effect size estimates are close to unbiased ***even when outcomes are auto-correlated***.
- Effect size estimates are typically more accurate than change-in-levels model, except when treatment effect is very close to zero.
- When outcomes are independent, the variance estimator performs poorly when treatment effects are close to zero.
- Variance is systematically under-estimated when outcomes are auto-correlated.
- For meta-analysis, robust variance estimation is recommended to account for inaccurate variance estimates.

Figure 3. Root mean square error of the effect size estimate for the gradual effects model and the change-in-levels model when outcomes are independent.

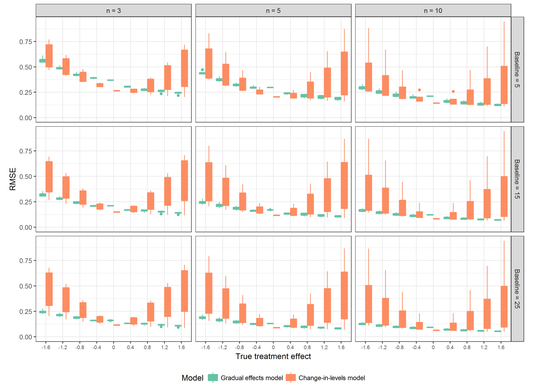


Figure 4. Relative bias of the treatment effect variance estimates from the gradual effects model when the outcomes are independent (left panel) or auto-correlated at φ = 0.4 (right panel).