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Editorial

Case Study and Single-Subject Research Investigations

*James P Dworkin-Valenti**

Department of Otolaryngology, Detroit Medical Center, USA

Editorial

Annals of Clinical Case Studies is an online-open access source of scientific information derived from case study investigations in numerous medical subspecialties, including allergy, cardiology, dentistry, dermatology, diabetes, gastroenterology, hematology, infectious diseases, infertility, nephrology, neurosurgery, ophthalmology, clinical neurology, nutrition and diabetes, obstetrics and gynecology, oncology, orthopedics and rheumatology, otolaryngology, pulmonology, and surgery. Within this broad scope of the journal, articles may represent "how I do it" case reports, unique case studies, the results of single-subject research designs, or rare condition case series. Each methodological format can produce informative findings that improve our understanding of a particular subject of interest. In the how I do it authors typically describe in detail a novel diagnostic or treatment approach, which they found to be superior to an accepted standard of care technique. This relatively informal paper allows the authors to elaborate on their personal experiences and successes.

A case study offers an intensive overview of a subject, group, event, or community with an interesting or complex condition that merits further informal review and discussion. Of note, most of Sigmund Freud's theories of psychological development and human nature were derived from detailed case study biographical investigations. For the most part, the case study design requires that researchers collect and evaluate in-depth descriptive data *via* interviews of the participant of interest and possibly friends, family members, and professional associates. Not infrequently, the case study requires researchers to scour medical charts and public records to gather sufficient data to report. It must be understood that in case study investigations cause-effect relationships are not usually established because in virtually all instances study variables are not discretely defined or manipulated to test specific a priori hypotheses; that is, standardized experimental research methods are not employed to demonstrate validity of the findings observed or obtained. Not with standing this limitation, result of case study investigations can be very valuable because they often extend current knowledge on various subjects of interest. Whereas a case study usually describes that which has already occurred (i.e., retrospective), on occasion it is designed to evaluate a dynamic situation or planned event (i.e., prospective). It could be argued that many case studies prove to be nothing more than elaborate "how I do it" publications.

A significant disadvantage of the case study methodology is that the information presented is extremely subjective because the qualitative data are derived from poorly controlled methods of collection; nuisance variables and author biases frequently exist to confound the findings and contaminate both internal and external validity. Case studies usually do not permit researchers to determine the causal relationships between specific variables or events.

The case series design is primarily an extension of the case study method, wherein (preferably) several pathological specimens or subjects with conditions that closely resemble one another are examined in detail. Medical journal editors are inclined to entertain such paper submissions if the set of subjects (or specimens) studied is a good representation of a unique, controversial, or poorly understood clinical population or pathological condition. Still, results of a case study or case series usually do not lead to unequivocal conclusions. Instead, they provoke food for thought or theoretical considerations for additional research or theory development.

A single-subject research design gathers quantitative and highly structured data from a single subject or group of single subjects. Unlike a case study this quasi-experimental method helps investigators comprehensively examine the possible causal relationship between specific interventions and behaviors of interest. Generalizability to larger populations (i.e., external validity) remains limited with this research design, and like the case study there is no control group for comparative analyses or support of the findings. Notwithstanding this inherent shortcoming, the single-subject investigation affords researchers control over the data collection process to demonstrate whether or not manipulation of an independent variable (i.e., therapy protocol) causes an expected change in the dependent variable (i.e., treatment target). Minimizing extraneous or nuisance variables is of paramount importance to ensuring internal validity of the anticipated cause-effect relationship between these two study variables. To achieve this objective, researchers must accumulate repeated measurements of treatment target behaviors to establish an accurate and consistent profile of the problem prior to the institution of the intended treatment. Whether the investigation involves only one subject or multiple subjects with similar difficulties, each subject is treated as a solo experiment instead of one trial in a larger experiment. That is, the investigation examines how each unique individual subject responds to the experimental factor; an especially helpful approach when researchers are interested in specific subsets of a particular demographic group. Subjects thus serve as their own controls throughout the experiment.

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***Corresponding author:** James P Valenti, Department of Otolaryngology, Head & Neck Surgery, Detroit Medical Center, Detroit, MI, USA, Tel: 313-966-9471; E-mail: voicedx@yahoo.com

In a traditional experimental group study design, often with a control subgroup, researchers usually must recruit a large number of eligible participants in order to demonstrate statistical power and significance of the results. This can be a very time consuming and difficult task. Comparatively, with the single-subject research design investigators can more quickly conduct their experiment because far fewer participants are required for data collection to commence. Naturally, the glaring downside of this design, as mentioned earlier, is the restricted ability to extrapolate positive results to the study population at large. Obtained findings only show how the individual subject responded to the intervention; external validity is lacking.

A single-subject research design is frequently used in experiments within applied fields of psychology and education. It is sometimes called a small-n design, wherein "n" represents the statistical symbol for the sample size; usually somewhere between two and 10 participants. For this reason especially, this type of experimental research often yields good internal validity. That is, little doubt whether the treatment administered was the cause of a subject's improvement. Although there are several different single-subject research designs, a couple of features are common to most of them. First, the dependent variable or treatment target is measured repeatedly over time at regular intervals. Second, the experiment is usually divided into distinct phases, wherein the subject is administered the independent variable (treatment) under only one specific condition per phase. Conditions of the experiment are usually defined by 3 capital letters (e.g., A, B, or C). Change from one condition to the next depends on the subject's behavior; the researcher typically waits until such behavior is fairly consistent (steady) over multiple observations before switching conditions. Thus, when the target behavior has reached a steady state any changes observed thereafter across treatment conditions can usually be easily detected.

As noted earlier, with the single-subject research design there is very little interest in comparing responses from different subjects who may be exposed to the same treatment paradigm. Rather, careful analysis of each subject separately is vital to the integrity of the study, not group performances. Reliability of the results depends not on group averages, standard deviations, and inferential statistical analyses of a large cross-sectional data base. Instead, determining reliability of findings in a single-subject research design is largely dependent upon the ability to demonstrate that a subject's responses to the intervention can be replicated over and over again by that individual. This research approach provides feedback quickly to the experimenter about whether and when the treatment rendered is effective; these answers are usually evident relatively soon within the intervention program. Variability in responses are observable daily, enabling real-time modifications to effect desired performances, unlike the rigors of large group designs that are not friendly to day-to-day adjustments to tweak responses of the subjects. That is not to say that the single-subject design is not rigorous. It is very much so, because the researcher uses regimented procedures to establish control over all experimental conditions in order to obtain desired stable and replicable behavior in response to a specific treatment condition. However, it should be noted that it not infrequently takes several months for complete study of a single subject under the various treatment conditions of interest. Acquiring stable baselines prior to administration of treatment is often very difficult, and behavior variability can be due to intrinsic as well as extrinsic factors that must first smooth out. Such stability can be arbitrarily defined by the researcher at the outset, but usually includes at least several days of testing to be demonstrated. The treatment experiment should not begin until a reasonable stable baseline of target behavior is established. Manipulations of intrinsic and/or extrinsic factors may be required to achieve baseline stability.

The two most common types of single-subject research methodologies are 1) Reversal, and 2) Multiple baseline designs. The reversal method is the most basic, often called an "ABA" design because of the 3 associated conditions of the experiment. In the first one, from left to right, "A" represents the stable baseline of an abnormal behavior, prior to the introduction of a planned treatment. This baseline phase could constitute the control condition. In the second one, after this steady state baseline is obtained, phase "B" begins. This is when the treatment is applied and positive or negative responses are graded over a period of time to demonstrate that an arbitrarily defined level of change is observed in the targeted behavior. In the third phase, once a plateau therapeutic effect is observed over this stable period of time, the treatment is withdrawn (i.e., reversed or taken away) to determine if any improvement worsens as a consequence. The researcher waits until the target behavior once again reaches a steady baseline state ("A"). If replication in the same subject and other subjects can be demonstrated following this method of data collection, conclusions about treatment efficacy would be strengthened.

If there are different kinds of treatment conditions, then the others are often called "C", "D", "E" and so on. It is noteworthy that AB designs are very weak because they cannot produce results that demonstrate unequivocally specific treatment effects; any number of unknown concurrent environmental conditions or events, not related to the treatment, may account for the improvements observed in the target behavior. Thus, the AB design does not permit denial of alternative explanations for positive treatment results because proper outcome controls were not employed in the experiment. To strengthen the hypothesis that it indeed was the treatment that caused the notable behavior improvement, a more powerful design can be employed for such purposes.

The "ABAB" approach is the most powerful design strategy to measure treatment impact. With this method, the researcher begins collecting stable baseline data ("A"), followed by introduction of the treatment for the first time ("B"), followed by withdrawal ("A") of the treatment to induce reversal of behavior toward the previously stable baseline level, followed by reintroduction of the treatment condition ("B") for the second time. Accordingly, this single-subject research methodology may allow the investigator to make stronger conclusions about treatment effects than is possible with the "ABA" design if each time the treatment is applied notable improvement over the baseline level is observed in the target behavior. In other words, the reversal strategy markedly increases the internal validity of the cause-effect treatment conclusions. Theoretically, to strengthen this argument even more, the researcher could employ an "ABABAB" design. However, if during the "ABAB" design the baseline level cannot be recovered after the treatment withdrawal phase (i.e., second "A"), or if such a reversal procedure would pose an ethical or harmful dilemma for the subject, a different methodological strategy, such as the multiple baseline design, may be necessary to measure treatment efficacy. This alternative will be discussed shortly.

Variations of the reversal design exist including a "C" phase that may augment the treatment being administered. This additional experimental condition could be strong praise or punishment for

each of the subject's performances; each can be altered in sequence to control for order or carryover effects. For example, a multiple treatment reversal design could follow an "ABCACB" sequence of experimental steps.

The multiple baseline design can serve as an alternative single-subject research methodology for the "ABAB" approach when it is not possible or ethical to withdraw a treatment to induce reversal to the baseline status. In one version of this option, a baseline is determined for each of several subjects followed by introduction of the treatment for each one; that is, multiple AB methods, but at a different time for each participant. The objective with this approach is to demonstrate that when the target behavior changes in a subject after administration of the treatment it is unlikely to be due to extraneous conditions. Such assurance is accomplished if multiple subjects, independently treated in the same way at different time intervals, respond similarly to the independent variable. When replication is demonstrated across the subject pool the treatment effect is extremely unlikely to be the result of coincidental experimental factors.

In another version of this design, multiple baselines are collected from a single subject but for different target behaviors (i.e., dependent variables); the specified treatment (i.e., independent variable) is then introduced for one target, and at a later date the same treatment is administered to effect change in another declared target. The intent of this method is to produce strong evidence that the treatment itself was very likely responsible for the notable change in the target behavior. If improvements are observed in two or more related target behaviors after treatment application at independent and disparate time intervals, researchers can with confidence suggest cause-effect experimental outcomes.

A third version of this design requires obtaining multiple baselines from the same subject but in different settings; for example, at home and in the clinic. The treatment would be administered at one setting first and at a later time at the other location. If the target behavior changes after the treatment in each of these settings, the data would support conclusions of cause-effect outcomes.

Interaction and alternating treatment designs may be used by researchers as extension methodologies of the basic ABAB design. In the first of these designs, the objective is to determine interactive or independent effects of two different treatments on a single target behavior in at least two subjects. In the second of these designs, the relative effectiveness of two different treatments (A or B) on a single target behavior in one subject is measured by alternately presenting these treatments according to a counter-balancing sequence. For example, ABBA *vs.* BAAB**,** administered at different time intervals to establish whether one treatment result is superior to the other, controlling for possible order effects *via* this counter-balancing technique.

Data analyses in single-subject research designs differ from group research methodologies. In the latter, researchers typically employ various parametric and nonparametric inferential statistical computations to determine both the significance of experimental findings and whether the results can be generalized to a larger representation of the study population. By contrast, single-subject research designs rely mostly on visual inspection of assorted data points represented on line graphs of one type or another. These plots enable researchers to identify patterns, slopes, and trends in behavior changes over specified intervention time intervals. Graph paper is usually used to display data points and dots so as to reduce errors or misinterpretations of subject performances. The vertical or y-axis displays the target behavior, during baseline and treatment measures. The horizontal or x-axis is used to indicate phases or conditions of the experiment, including treatment sessions and trials over specified periods of time. Use of certain statistical measures, such as percentages, means, and standard deviations can be employed in single-subject research to strengthen conclusions about treatment outcomes. Quantitative statistical computations such as a t-test or analysis of variance are not infrequently applied to analyze the data obtained and to determine the actual significance level of the treatment responses. Occasionally, when data or observations collected repeatedly from two or more subjects are non-independent, autocorrelation analyses can be used to compare data collected at the baseline phase and at each of the experimental intervention phases. Consultation with a bio-statistician is recommended for guidance about the most appropriate analysis measures to utilize to demonstrate possible cause-effect results for the types of data gathered.

Summarily, the list below represents 10 essential methodological rules of thumb that authors should follow at the outset of a case study or single-subject research investigation and when preparing the manuscript for submission to a scientific journal for publication consideration:

The case presentation must be grounded in a scholarly evidence base, both for its motivation and its interpretation.

1. The scholarly literature base and theoretical framework that relates to the case must be comprehensively examined in the Introduction section of the paper.
2. The literature review must support the need for the case report, case series, or single-subject research experiment.
3. The Methods followed for data collection must be described in detail; they should include the various techniques employed to a) reduce examiner biases and nuisance variables, b) ensure reliability of judges' impressions, c) strengthen internal validity, and d) support all conclusions drawn.
4. Descriptions of the medical, personal, and psychological background history of the case(s) or series of subjects must be presented in detail.
5. Essentially, the case description must be sufficiently detailed to enable other interested researchers to replicate the results with similar cases in their practices.
6. All evaluations that are conducted and all treatments that are rendered must be explained in great detail so that others can replicate the study.
7. Supportive figures, pathology slides, and tables should be included to augment and substantiate the narrative.
8. The Discussion section must seamlessly connect the results obtained to the background literature on the topic of the case investigation, for distinctions, similarities, generalizability to a larger population sample, and future research implications.
9. Finally, no case study, case series, or single-subject research investigation should be considered ready for submission to a journal without definitive information relative to the final outcome; readers should not be left wondering about the ultimate disposition of the subject (s) at the end of the manuscript.