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Scientific case research in complementary and alternative medicine—A review

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Summary The description of individual cases is probably the most important didactic tool for teaching in medicine, especially in complementary and alternative medicine (CAM). However, only very rarely is the information provided in traditional single case reports sufficient to answer scientific questions. These reports typically are demonstrations of the solution to a clinical problem. To contribute to scientific discussion single-case research must ask critical questions whose answers are open. Two fundamental questions are: (1) Is my observation reliable? (2) Which factors, other than my treatment, may explain the observed outcome? In this review we will give an introduction to single-case research, as well as present and explain single-case designs as a tool for research and discuss their relevance and applicability for clinical practice in CAM. This review deals exclusively with single-case research on treatment effects and covers observational single-case studies, progressive and repetitive experimental single case designs, n-of-1-RCT, multiple baseline design, best case series and meta-analysis of single-case studies.

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Introduction

Why are single-case studies needed? The most important didactic tool for teaching in medicine is probably the description of individual cases. Listening to a patient's narrative of his/her disease, including their personal pattern of signs and symptoms and what happened once they received treatment is remarkable. Proponents of CAM treatments typically refer to reports of individual cases to highlight the benefits of their method. But why is it that, in general, case reports play a minor role in providing evidence to scientifically prove that a treatment is effective?

Most case reports in CAM today are retrospective accounts of an impressive case as perceived and documented by a therapist. They serve more as a narrative to demonstrate a problem or the solution to a problem in clinical practice and can be considered very important for teaching sessions, as a didactic tool, to help students remember key information. They should be exemplary and encapsulate all the relevant clinical issues. A well told story of a case will stick in one's memory and the crucial points can be called upon if a similar patient appears for treatment in future clinical practice. Sometimes observations in single cases can even generate new hypotheses or identify new treatment options.

However, most case reports cannot be considered systematic research. Instead of ''demonstrating'' something, empirical research tries to answer open questions. The methods used must be transparent and stand a critical evaluation. For example, imagine a report by a therapist who is convinced to have a fabulous cure for a patient with advanced cancer. Even if one is only slightly sceptical, numerous questions will immediately arise: Did the patient truly suffer from this specific type of advanced cancer? Was the patient truly cured? If so, did he/she receive other treatments which might have contributed to the cure? Could it have been a spontaneous remission? Only very rarely is the information provided in traditional single case reports sufficient to answer such questions. To be considered research, critical questions must be addressed in a case report or a single-case study. In our view, the two most fundamental questions are: (1) Is my observation reliable? (2) Which factors other than my treatment may explain the observed outcome?

Surprisingly, the number of systematic single-case studies published in the field of CAM is very small. This is a pity, in our opinion, because such research could contribute considerably to the critical evaluation of CAM therapies and CAM research may benefit from improved understanding of benefits, limitations and utility of single-cases.

Furthermore, although the workload associated with high-quality single-case studies must not be underestimated, they are probably the most feasible research tool for the busy practitioner. In this review we want to give an introduction to single-case research, to present and explain single-case designs as a tool for research and discuss their relevance and applicability for the clinical practice.

This article exclusively addresses single-case research on treatment effects. The aim of all such research is to provide evidence for causalities during the therapeutic process. We do not cover etiological, prognostic, diagnostic or procedural research. Throughout this article we will use the term ''case report'' if we refer to retrospective analyses of single patients and the term ''single-case study'' if there is at least some prospective element to the study.

Quantitative and qualitative approaches to single-case research

While single-case research is applied in diverse fields, such as medicine, education, rehabilitation, sport and athletic performance, it has particularly strong roots in psychology. Several textbooks summarizing methodological approaches are available (e.g. Refs. 1—3). These textbooks focus mainly on experimental single-case studies (see below) with graphical and statistical analyses. In such studies the influence of an ''independent variable'' (e.g. the treatment vs. a control condition) on a predefined outcome (e.g. blood pressure) is investigated. The outcome is measured repeatedly (e.g. daily over 12 weeks). The findings are then displayed using graphs. Specific statistical analyses are applied to test whether changes in outcomes exceed those expected by chance.[4] This approach to single-case research is strongly quantitatively orientated and the principles in drawing inferences regarding causality do not differ greatly from research performed with groups of individuals.

A second important field of research are qualitative case studies. This type of research is often carried out by medical anthropologists or social scientists. Especially when a researcher is investigating a field of research where little is known, a qualitative case study or case series might be a good approach to enter the field, to collect data and develop scientific hypotheses and theories. A qualitative method