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Integrative medicine case reports: A clinicians' guide to publication

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ABSTRACT

Case reports have been a valuable method of informing medical practice for as long as medicine has been practised. Many original observations, novel diagnostic and therapeutic approaches, unusual, new or uncommon diseases and complications of medical treatment were first identified and published as case reports. Despite their importance, and contemporary trends supporting their further use, publishing case reports can be a difficult task for many clinicians and researchers. Preparing a case report for publication can be both a professionally and personally rewarding endeavour for clinicians. This article describes practical and academic insights into writing a case report for publication.

1. Introduction

Case reports have informed medical practice for as long as medicine has been practised. Derided by some, but adored by others, the published case report has had a long and chequered history in evidence-based medicine. In the age of the clinical trial and evidence-based practice, published case reports continue to capture and describe important scientific and clinical observations that may be missed or undetected in 'higher hierarchy' designs such as clinical trials. Additionally case reports can provide important patient-centred clinical insight that may inform the individualised nature of contemporary patient care. Case reports can generate hypotheses for future clinical studies, guide the personalisation of treatments in clinical practice, and (particularly useful in integrative medicine) help to evaluate systems-oriented approaches to healthcare [1].

2. Why case reports matter

History demonstrates the importance of published case reports in modern medicine. HIV was brought to the mainstream medical world's notice not through exhaustive epidemiological monitoring, for example, but through publication of an influential case report of "extensively disseminated Kaposi's sarcoma in a young homosexual man" [2]. Case reports have also led to new advances in knowledge of existing diseases. Publication of a case of rabies encephalitis eight years after exposure defied previous accepted norms of rabies incubation (thought to be one to six months) and suggested the existence of a novel, slowly proliferating subtype previously rejected by neurologists [3]. New and novel treatments have also been identified through case reports – propranolol as a treatment for infantile haemangioma, for example, began as a case observation of nine children [4].

Case reports can also point to unknown risks or demonstrate regulatory or practice failures. Our understanding of the relationship between thalidomide and congenital abnormalities began with case reports [5]. In integrative medicine, case reports have often been focused on highlighting the risks in what is sometimes perceived by the public as a benign and harmless. The publication of a case report of accidental death from selenium overdose, for example, highlighted the risks associated with patient use of potentially safe nutritional therapeutics without clinician oversight or advice [6]. The uncovering of peripheral neuropathy due to undisclosed pyridoxine use, which confounded medical specialists treating a British woman for nearly ten years and was immediately resolved upon cessation of her supplements, highlighted the important clinical impact of not asking patients about their complementary medicine use (and as such probably reflects far worse on her attending physicians than her supplements) [7]. Case reports of missed diagnoses when complementary and integrative

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practitioners ignore conventional medical treatment [8,9] have been used to highlight current regulatory, practical or training deficiencies in clinical practice.

Whilst these 'negative' case reports are essential for ensuring integrative medical care is delivered in a safe and effective manner, they do little to inform the evidence around clinical practice in a positive sense, nor add to the evidence base for new and potentially novel treatments. For this to occur, the onus is one the integrative medical community itself to engage in reporting both the notable failures and successes of integrative medical treatment. In doing so, the integrative medicine community benefits by developing increasing research capacity within the integrative medicine professions, as well developing a better understanding of specific integrative medicine interventions and techniques.

3. Building research capacity in integrative medicine

Scholarly writing, as done through publication of case reports, can offer valuable learning experiences for clinicians, offering insights into their own practice that result in improved clinical care for their patients [10]. The process of writing also obliges mental and practical discipline. The peer-review process undertaken in submitting a case-report may offer valuable feedback that provides insights to clinicians beyond their training or clinical practice experiences. Partaking in scholarly writing activity can also help develop the field of integrative medicine, by increasing the research capacity of the integrative medical field, which lags other fields in ability to fully engage with research activity [11]. Without clinician engagement in the research process, research is unlikely to accurately reflect the realities of clinical integrative medical practice; however, if clinicians do not engage with research critically, the fruits of their efforts are unlikely to be influential in an evidence-based paradigm [12].

For case reports to appropriately inform evidence-based practice, they must be presented in a scientifically rigorous manner. Just as good case studies can inform good medical practice, poor case studies can lead not only to rejection, but in some cases actively deny integration of beneficial treatments or allow the proliferation of non-beneficial treatments. Case reports of hepatoxicity related to kava ingestion (*Piper mythesticum*) which were used to support bans of the substance in numerous countries, for example, often failed to differentiate between solvent-base and aqueous-based extractions or negated to acknowledge the presence of other known hepatotoxic agents in kava combination products [13]. The result was premature removal from the market of an effective and safe therapy for a condition in which few other equally safe and effective therapies existed. As case reports form the lowest level of the evidence hierarchy, notable findings can often be used to argue for, and implement, larger studies evaluating these findings.

As such, case reports may have impact 'higher up the evidence chain'. Good case reports can inform larger studies, promote the uptake of effective novel therapies and offer clinical insights into rare or uncommon conditions. Poorly written case studies (for example, those that include confounders such as additional treatment – either by other parties or self-prescribed by the patient – which are undisclosed in the report), however, can lead clinicians and researchers down the proverbial 'garden path', expending valuable time on resources trying to replicate results that may be completely unachievable. For this reason, there have been recent attempts to develop quality assurance measures for case reporting, and these are being adopted by *Advances in Integrative Medicine*.

4. Standardisation of case report publication: the CARE guidelines

Although the case report itself is experiencing a renaissance in clinician and researcher support, lack of standardisation and multiple guidelines have led to case reports historically being of variable quality and clinical relevance, and have made it difficult for the findings of case reports to be effectively used as part of the foundation of evidence-based medicine [14,15]. To help facilitate the important role of case studies in informing evidence-base medicine, consensus-based clinical guidelines have been developed to address the important issue of standardisation of case reporting. Such guidelines have already been developed for other clinical research designs including clinical trials (CONSORT: COnsolidated Standards of Reporting Trials), observational studies (STROBE: STrengthening the Reporting of Observational studies in Epidemiology) and systematic reviews and meta-analyses (PRISMA: Preferred Reporting Items for Systematic reviews and Meta-Analyses).

These guidelines, the CARE (CAse REporting) guidelines [16], have been adopted by leading international journals, and provide a framework that supports transparency and accuracy in the publication of case reports and the reporting of information from clinical encounters. Like the guidelines for other clinical research designs mentioned previously, these guidelines now form the international standards for medical literature (further information can be found on the EQUATOR Network website: http://www.equator-network.org/). Not only does this standardisation result in case studies that are most likely to provide data that informs clinical practice guidelines and provide early signals of effectiveness, harms and costs, but standardisation can also mean that case reports from all journals utilising these guidelines can be analysed either individually or part of a much larger database of reports. *Advances in Integrative Medicine* also uses the CARE guidelines for case reporting. Details, including templates and examples of case reports using the CARE guidelines can be found on a dedicated website: http://www.care-statement.org/.

5. Other discipline-specific guidelines

In addition to general publication guidelines, individual integrative medicine disciplines bring with them their own unique challenges. For example, in addition to general case reporting guidelines, it would be expected that acupuncture case reports also comply with the acupuncture-specific STRICTA publication guidelines, which detail acupuncture-specific requirements such as how to report acupuncture rationale, needling technique and practitioner background [17]. For this reason, in addition to the generic case reporting guidelines described in this article, it is encouraged that authors also familiarise themselves with and seek guidance from discipline-specific guidelines. These have been developed for (medical) acupuncture [18], Chinese medicine [19], chiropractic [20], herbal medicine [21], massage [22], naturopathic medicine [23], and pharmacy [24], among others.

6. What should be reported?

Advances in Integrative Medicine receives many case reports that do not comply with the guidelines above, and as such must reject the bulk of them before even sending out for review. Others have been sent for review, but have not been recommended for publication due to problems associated with not meeting these guidelines sufficiently. The high rejection rate for case reports by this journal, despite this journal's commitment to publication of notable case reports, served as the impetus for outlining requirements in this article, and the development of further criteria on our website. However, even when case reports do