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Open-access electronic case report journals: the rationale for case report guidelines

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1. Introduction

Case reports serve three primary functions: generating hypotheses for verification in subsequent longitudinal observational studies and clinical trials, documenting rare or unusual phenomena where obtaining further evidence is not feasible, and providing clinical stories that help health care providers remember important lessons. Vandembroucke [1] previously discussed these roles in depth, describing how the use of deduction, induction, and serendipity could produce case reports that lead to more formal investigations, case report strength of evidence, and the aggregation of multiple case series into meta-analyses to generate meaningful conclusions. The latter two observations increasingly have been viewed with skepticism, partly because of the rise of evidence-based medicine (EBM) in the early 1990s. EBM strives to integrate the best available published research with both clinical expertise and patient values and circumstances [2,3]. Case report findings are not generalizable, do not address causal inference or explanatory mechanisms, and emphasize low-probability events [4,5]. Systematic reviews or meta-analyses of multiple case reports or case series add numerical weight, which can improve the chances of detecting unusual associations. However, pooling data does not in itself allow a rigorous assessment of causation when the source material is composed solely of multiple case reports lacking longitudinal data. In addition, sufficiently complex diseases may have heterogeneous clinical presentations. Simply pooling cases that ostensibly represent the “same” disease can sacrifice sensitivity to increase specificity, while in the process deemphasizing salient details of each case [6].

The EBM movement consequently tends to categorize case reports as weak evidence, and we agree to an extent. However, that does not mean that they are of little importance. Their role in uncovering important phenomena originates in the ancient past: Hippocrates’

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Aphorisms, the Edwin Smith Papyrus, and the *Yellow Emperor's Classic of Medicine* are all compilations of case reports and anecdotes that are among the first written documentation of medical concerns [7]. In contemporary times, the first article of the inaugural issue of the *New England Journal of Medicine* 200 years ago was a descriptive treatise on a series of patients with angina pectoris [8], and five of the 51 articles in *JAMA's* 1985 centennial compendium *Landmark Articles in Medicine* were case reports [9].

The educational value of well-written case reports is also underappreciated. The human brain is particularly good at learning and remembering through examples and anecdotes, and case reports can offer valuable and memorable examples of how a disease might present or how to avoid certain complications when doing new procedures [1,7,10,11]. Health care providers can benefit greatly from discussing patient stories, which can be more effective and inspiring than impersonal statistical abstractions. Recognizing that medicine is as much a story-based narrative process as a scientific one can enhance empathy with patients and provide perspective [12]. For addressing clinical conundrums, case reports can help generate hypotheses, highlight ethical pitfalls, and bring to mind diagnoses that health care providers might have otherwise overlooked.

Herein, we will discuss two additional phenomena that have further obscured the already debated role of case reporting in modern medical practice. First, the recent rapid growth of open-access online-only journals focused on case reports has lowered authorship and editorial barriers to publishing. Second, the continued lack of publishing guidelines for case reports has led to variation in quality and thematic presentation across journals and clinical disciplines. Consequently, clinicians intending to incorporate useful findings from case reports into their practices face a daunting challenge. We outline current case report publication trends in the later section and provide suggestions for quality improvement in case report writing and editing that may help strengthen their clinical, scientific, and educational value for health care providers.

2. Case report publication trends

An April 2012, general search for “case report” on PubMed and EMBASE yielded about 1.6 million and 2 million hits, respectively. These represent about 8% of all published records in both databases since their inception [13,14]. The total number of new articles classified as case reports for each year from 2000 to 2010 is shown in Fig. 1. Annual case report volume increased from 42,439 publications on PubMed and 49,918 on EMBASE with MEDLINE in 2000 to 61,689 and 72,388, respectively, in 2010, a 45% increase in both databases. Among clinical specialties, the proportion of published case reports and series varies widely. Within high-impact journals of internal medicine and general surgery, the proportion of case reports was about 10–12% during the 1992–1994 and 1996–1998 time intervals [15]. In more specialized fields, case reports may comprise as much as 88% of all publications (Table 1).

3. The online case report publishing movement: risk vs. reward

BioMed Central and the BMJ Publishing Group (publishers of the influential *British Medical Journal*) are two institutions that have pioneered the online distribution model for medical publishing, using case reports as the exclusive foundation of new subsidiary journals. The first case report-only journal, *Journal of Medical Case Reports*, was launched by BioMed Central in January 2007 [16]. In 2008, BioMed Central established a second case report-only periodical, *Cases Journal* [17]. The same year, the BMJ opened the *BMJ Case Reports* Web site, which publishes articles online on a continuous basis. Each of these three journals offer novel perspectives on case reports—for example, the *Cases Journal* will accept submissions directly from patients. These journals hearken back to a very traditional style of learning, one of peer discourse and professional communication of cases both

routine and unusual. However, the visibility of these contemporary pioneer case report-only outlets has triggered a flood of similarly themed imitators. An April 2012 Google search for “case report journal” revealed 19 separate case report-only journals in the first 30 hits alone, nearly all of which are open-access and online-only journals.

The open-access, electronic-based, case report-only publishing model has some advantages. First, many of these publishers, including the BMJ, emphasize high acceptance rates or rapid editorial review in their advertising to potential authors [18–20]. Second, for the busy clinician with little familiarity with research methodology, the case report is the simplest method of describing clinical experiences that potentially serve as “gateway” projects to more in-depth future investigation. In areas of the world with fewer resources, that is, low- and middle-income countries (LMICs), the infrastructure and expertise to perform clinical trials and other complex research simply may not be available. Hence, the case report has been promoted as a means of circulating potentially useful clinical knowledge within professional communities in LMICs. Furthermore, the open-access case report journal is an instrument for supplying resource-poor areas with medical knowledge from around the rest of the world [21].

However, these advantages must be weighed against several important problems with the proliferation of open-access case report journals. First, prior research has outlined concerns regarding both editorial and authorship quality standards in case reporting. In 2004, Sorinola et al. [22] found that among 163 journals that published case reports, editorial guidelines tended to emphasize technical issues such as word and page limits, abstract structure, and keywords more often than actual case report content. Fifty-five percent of the journals required an instructive or teaching point, but only 6% required hypothesis generation as a reason to submit a case report. Richason et al. [5] evaluated 1,316 case reports and series in four major peer-reviewed emergency medicine journals from 2000 to 2005. They found that more than half of the publications were missing critical information such as active medications, cointerventions, and adverse effects of the primary intervention. Furthermore, less than one-third provided alternative explanations for favorable outcomes or generalizability of results. These studies suggest that case reports are of inconsistent quality, and readers may fail to gain anything meaningful from them and may draw inappropriate conclusions as well.

The second problem is practical in nature. The absolute number of case reports entering PubMed and EMBASE/MEDLINE now exceeds 60,000 annually, 50% more than the figure cited by Rosselli and Otero [23] one decade ago, and the actual figure could be even higher because many of the new case report journals are not indexed within these libraries. An explosion of case reports might drown key sentinel events in a sea of careless publishing as readers will be challenged attempting to locate articles of specific clinical interest out of the millions of case reports in the medical literature. Furthermore, for journals aiming to improve their visibility through increasing impact factors, it has been speculated that case reports are greatly read but rarely referenced, thus having little appreciable impact on citation figures [24].

Finally, providing free access to readers is not without cost. Nearly all journals charge authorship fees, often as high as thousands of US dollars, to make a given article freely available to the public. In the case of open-access electronic-only journals, authorship fees are nearly always mandatory, even if there are no printing or peer-review costs [25]. For writers with substantial resources, the cost may be nominal, but for those in LMICs, this fee may be a barrier to publishing their own case reports, if not preventing them from reading others' manuscripts.

4. Suggestions for improving case report standards

We propose that the optimal case report should follow the format of an “old school” Grand Rounds, including putting the case into appropriate perspective from a clinical and/or research audience. If the impetus for the case report is educational, then the evidence supporting it as an illustrative case must be well documented and articulated. We present in the later section a potential checklist for high-quality case reporting, based on suggestions from prior publications [5,26,27] and welcome contributions from others seeking to refine or expand on it (Table 2).

4.1. Title and abstract

Like all reports of medical findings, a case report should clearly state its purpose, including whether it is illustrative/educational or a report of a phenomenon meriting further investigation. Abstracts should also contain other information useful to a busy clinician who wishes to quickly judge whether the article is relevant to their clinical practice, such as the diagnostic, prognostic, or interventional topic to be covered, and the specific lesson to be learned from the case when applicable.

4.2. Introduction

Many open-access journals have relaxed standards for case reports [21]. We believe this premise is flawed. This increases the risk of trivial or misleading reports and does not show adequate respect for the time of the journal’s readership. Mundane observations should be left to textbooks, where context experts and editors can filter available evidence more efficiently [28]. Adequate background material is necessary to allow the reader to understand the context in which the unique findings of the case reports are being presented. A clear purpose and rationale for the article should be provided.

4.3. Case report

The components of this section are a slight modification of recommendations by Richason et al. [5], whose study documented multiple critical absences of information that would better inform the reader as to the seriousness of the patient’s condition, the scope of observation or intervention, and inherent limitations of the cases being described.

4.4. Discussion

The contents of the discussion depend on the primary purpose of the case report. If primarily educational, providing clinical context is essential, including referencing the literature heavily to justify key takeaway points. Those aspects of the case that are typical and those that are less so should be made clear. If the report emphasizes a phenomenon that merits further study, the rationale for why the case warrants attention should be made. Although this argument will have a qualitative component by necessity, the logic for why this is unlikely to be simply a chance or expected event should be explicit. In particular, the significance of the observation and the speculative value of the case report are relevant. At the same time, a disclaimer should be made that case reports or series examine unique individuals and cannot necessarily be generalized to the broader population or even the reader’s patient population.

4.5. Conclusion

The conclusion should concisely summarize the primary point of the case report in one to two sentences.

5. Further considerations for case report guidelines

If case reports are to be respectable and useful instruments for improving patient care and advancing research, improving their content and presentation quality is important, and we hope that reporting guidelines might be one method of achieving this goal [5,29]. Although scientific writing per se is not one of the six core competencies of the Accreditation Council for Graduate Medical Education, communication is. As case reports are in many instances simply a written conveyance of a patient's presentation and management, poorly written case reports may well reflect a broader inability to communicate effectively with colleagues [30]. Subpar writing has even been associated with increased litigation and reduced compliance from patients [31,32].

Case reports are a form of scientific writing that does not require the same investment as most original research and therefore could be a useful tool for teaching written communication and critical thinking. Evidence suggests that formal graduate and postgraduate medical training does not adequately equip health care providers with scientific writing skills [30,33]. This has been attributed to lack of training opportunities or mentorship [34], decreased financial support and time for medical writing coursework [35], and a lack of incentive to publish in mainstream journals [36]. In an increasingly globalized health care provider community where English is considered the *lingua franca* of medicine, non-native English speakers may also benefit from training in scientific writing [37]. A set of written case report guidelines additionally would be of value not only to countries with well-established medical education infrastructure but also to LMICs with fewer resources and infrastructure for more sophisticated research methods. In these communities, practical dissemination of case reports may be a supplemental but effective method of teaching. Standardized case report language would also facilitate computerized database searching, indexing, and telecommunications.

Given the rapid technological advances in computing and telecommunications, open-access online case report journals are unlikely to decline in relevance. We should promote the role of these journals as outlets for educational case reports. The guidelines we have outlined can serve as a starting point for collaboration with organizations like the EQUATOR Network, an international organization that has been promoting higher accurate research reporting quality [38]. These organizations have the capacity to work with electronic publishers of case reports in the role of an educational clearinghouse, and they may also have the leverage to address the issue of high authorship costs, particularly for LMICs. These strategies may help restore the case report to a position of importance in medical practice and the research community.

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What is new?

- The rise of open-access online journals dedicated specifically to case reports has facilitated the production and dissemination of these articles worldwide. Case reports account for more than 60,000 new articles annually in the PubMed and EMBASE databases and comprise up to 88% of publications within selected clinical specialties.
- The primary advantage to the new case report publishing model is relative ease of manuscript submission, particularly for health care providers without the expertise or resources to conduct methodologically advanced research. However, the lack of case report writing guidelines has resulted in inconsistent case report quality.
- This article suggests formal publishing standards that can improve the overall quality of case reports and help health care providers use case reports more effectively in the clinical and research settings. These standards may also be valuable as educational tools for general medical writing, a critical skill for health care providers.

Fig. 1.
New case reports published in major medical databases.

Table 1

Proportion of case reports and series in the published literature of selected medical and surgical specialties

Specialty	Citation	Years evaluated	Estimated proportion of case reports and/or case series (%)	Source of data
Anesthesiology	Bould et al. [39]	2007, 2008	10	9,684 articles in 18 journals
Dermatology	Akerman et al. [40]	1998–2007	66–88	7,570 articles in 36 journals
	Mimouni et al. [41]	1993–2007	31–51	17,925 articles in MEDLINE
Emergency medicine	Henderson et al. [15]	1992–1994	26	2,013 articles in four journals
Facial plastic surgery	Xu et al. [42]	1999, 2002, 2005, 2009	62–65	975 articles in five journals
Hand and wrist surgery	Ahn et al. [43]	1988–2007	42	3,457 articles in seven journals
Neurosurgery	Hauptman et al. [44]	1996–2009	31	53,425 articles in 22 journals
Ophthalmology	Kumar et al. [45]	2005–2009	29	12,426 articles in 20 journals
Otolaryngology	Wasserman et al. [46]	1993, 1998, 2003	55–61	2,584 articles in four journals

Table 2

Recommended checklist for reporting and publishing case reports

Section/topic	Item no.	Checklist item
Title and abstract		
Title and abstract	1 ^a	<ul style="list-style-type: none"> Identify case report as <i>observational</i> or <i>interventional</i> Observational case reports can be subcategorized as <i>diagnostic</i> or <i>prognostic</i> Interventional case reports can be subcategorized as <i>pharmacological</i>, <i>procedural</i>, <i>behavioral</i>, <i>miscellaneous</i>, or <i>multi-interventional</i>
• Introduction		
Background	2 ^a	<ul style="list-style-type: none"> Describe relevant background information on medical observation or technique being described
Rationale	3	<ul style="list-style-type: none"> Describe reasons for publication: research hypothesis generation, education, or rare/unusual finding of clinical significance
• Case report		
Subject(s)	4	<ul style="list-style-type: none"> Describe patient demographics, including age, gender, and race/ethnicity State chief complaint and observation and/or intervention to be discussed
Patient history	5	<ul style="list-style-type: none"> List recent medical history, pertinent medical comorbidities, and social history Describe previously attempted interventions, if applicable
Medications and allergies	6	<ul style="list-style-type: none"> List pertinent medications and allergies
Physical examination	7a	<ul style="list-style-type: none"> Describe pertinent physical examination findings, symptoms, and signs
	7b ^a	<ul style="list-style-type: none"> If <i>observational</i> case report, clearly state key diagnostic or prognostic feature(s) that distinguish this case from previous ones
Intervention	8a ^a	<ul style="list-style-type: none"> If <i>pharmacological</i> case report, describe route and dosage, as well as side effects if present
	8b ^a	<ul style="list-style-type: none"> If <i>procedural</i> case report, describe surgical technique or procedure in detail, as well as complications if present. Diagrams are recommended
	8c ^a	<ul style="list-style-type: none"> If <i>behavioral</i> case report, describe type and duration of psychiatric or psychological intervention
	8d ^a	<ul style="list-style-type: none"> If <i>miscellaneous</i> case report, describe the intervention in detail, as well as complications if present
Outcomes	9	<ul style="list-style-type: none"> Describe time frame of observations and clinically relevant outcomes, including time to recovery or death
• Discussion		
Relevance of publication	10a ^a	<ul style="list-style-type: none"> If <i>observational</i> case report, describe clinical importance of findings, for example, fatal drug reaction or interaction, warning signs of impending adverse outcome, or unquestionably unique disease findings Describe potential alternative explanations for clinical findings

Section/topic	Item no.	Checklist item
		<ul style="list-style-type: none"> Describe feasible avenues for further research
	10b ^a	<ul style="list-style-type: none"> If <i>interventional</i> case report, briefly list or describe alternative treatment modalities that were considered List hypotheses for success or failure of intervention Describe feasible avenues for further research
Limitations	11	<ul style="list-style-type: none"> Insert statement regarding limitations of findings and difficulty in generalizing outcomes
• Conclusion		
Conclusion	12 ^a	<ul style="list-style-type: none"> Succinctly summarize key observation or technique

^aItems labeled “a” are items whose inclusion is strongly recommended, whereas unlabeled items should be given considerable flexibility in interpretation and inclusion. Depending on the type of case report, not all items are applicable.