

Capturing zebras: what to do with a reportable case

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Abstract

WE HAD ATTEMPTED TO PUBLISH A REPORT OF AN UNUSUAL manifestation of an uncommon disease discovered at autopsy. The case report was not accepted for publication because we had failed to order a highly specific test that would have unequivocally confirmed the diagnosis we entertained and subsequently wrote about. After much deliberation about this case and an extensive review of the literature, we now believe that there is a preferred approach to dealing with a reportable case. Because physicians seldom encounter cases that are reportable, we felt that sharing the important lessons we learned from our missed opportunity might prove helpful and encourage those who are considering preparing a case report for publication.

Always note and record the unusual... Publish it.
Place it on permanent record as a short, concise note.
Such communications are always of value.
— Sir William Osler¹

The 4 kinds of clinical cases that lead to case reports are (a) the unique case that appears to represent a newly described syndrome or disease, (b) the case with an unexpected association of 2 diseases that may represent a causal relation, (c) the “outlier” case representing a variation from the expected pattern and (d) the case with a surprising evolution that suggests a therapeutic or adverse drug effect.² Although infrequent, when these cases do present to physicians they are still often not reported because of a lack of recognition of their uniqueness, a misdiagnosis or simply a lack of thought or desire to disseminate the information.

Case reports appear to be the perfect match for house staff, who are required to produce scholarly work during their training,³ since they encounter a high volume of patients, some of whom may have reportable findings. The completion of a case report is often seen by house staff as easier and less time-consuming than other forms of scholarly work.^{4,5}

In reviewing the literature on case reports, we did not find any materials that highlighted tips and pointers or that stressed the pitfalls to avoid in order to write a strong case report. Although the care of the patient must remain the primary consideration, the purpose of this article is to present steps that should be considered when one encounters a potentially reportable case.

Steps to improve the chances that a report will be published

1. *Read, read and then read some more.* When a case comes along that is potentially reportable, it is crucial to read as much as possible about the condition. In addition to a broad literature search into the given topic area, we recommend another search combining the disease in question with the medical subject heading “case report” across a large database (e.g., MEDLINE from 1966 to the present). The reasons for this research are several. First, one needs to ascertain whether the aspect of the case that makes it seem reportable has been previously described and, if so, how many times and under what circumstances.⁶ Second, it is important to find out the specific diagnostic criteria required to confirm the diagnosis, and the standards of care in treating the disease. Third, reading published case reports in the same topic area will help in the prep-

Review

Synthèse

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- aration of the manuscript. It is important not to rely solely on consultants at the expense of thoroughly reviewing the literature.
2. *Order the appropriate tests to confirm the diagnosis.* It is important to order all of the medically appropriate diagnostic tests that will be required for publishing the case report. Factors such as medical necessity, patient preference and cost need to be considered. Financial liability for tests obtained primarily for the purpose of reporting the case may need to be determined.
 3. *Obtain informed consent.* Informed consent refers to an encounter between the physician and patient characterized by “mutual participation, respect, and shared decision making.”^{7,8} Written informed consent must be obtained for nonessential tests (those that do not directly affect patient care but are needed to write the report) and obviously for tests that involve risks to the patient (e.g., organ biopsy). In the latter situations, the risks to the patient and the potential lack of immediate clinical benefit of the results should be clearly documented in the patient record. Written informed consent need also be acquired for the eventual publication of the patient’s case summary.
 4. *Maintain patient confidentiality.* Take precautions to protect the anonymity of the patient described in the case report. For example, delete the patient’s initials, mask his or her eyes in photographs and remove telling details from the history. The International Committee of Medical Journal Editors urges authors to omit identifying details if they are not essential but to avoid altering or falsifying information to attain anonymity.⁹ Because the masking manoeuvres sometimes fail, it is crucial to obtain informed consent.¹⁰⁻¹²
 5. *Involve consultants early.* Consultants can be helpful in rare, unusual cases. Not only might they be able to add to the literature search in helping to determine whether the case in question is reportable, they might also be able to provide suggestions for the work-up and patient management. One should not simply consult whoever is on service for the month. Instead, ask for input from the specialist in the hospital division with the most knowledge and interest in the disease in question. If there are no local experts, we recommend that authors contact an expert outside of their institution (e.g., the author of a textbook chapter or review article on the disease in question). Likewise, it may be necessary to obtain a second opinion for any pathological specimen or to seek a pathologist with a special interest in the topic area.
 6. *Request an autopsy.* Autopsies are important to confirm antemortem findings, to provide insight into the extent of the disease (staging) and to discover new elements that may not have been clinically apparent. Autopsy examinations can be both helpful and humbling to clinicians, and they may provide guidance to the management of future patients who present with similar signs or symptoms.¹³⁻¹⁵
 7. *Save blood samples.* Whether the patient dies, moves away or is lost to follow-up, saving several vials of blood, which can be used for additional testing, is a reasonable suggestion. This practice can prove invaluable when a journal’s reviewers request specific laboratory tests that were not performed originally. Saving a blood sample may be most useful when writing a case report about a condition that is outside the authors’ specialty area and for which they are thus unfamiliar with the laboratory test results normally reported, or when access to further blood samples is unlikely. Authors should check with their institution’s laboratory about the requirements and allowances for freezing blood samples. Samples should be stored only if the patient or their representative has given consent.
 8. *Discuss the case report with the editorial staff of the journal to which you intend to submit.* An important function of the editorial staff is to help authors submit the best work possible. The “information for authors” pages often yield limited instructions for preparing a case report. Communication with the editorial staff early in the writing process about the format,¹⁶⁻¹⁸ the backlog of accepted case reports awaiting publication, the proportion of submitted case reports that get accepted and, most important, the journal’s interest in the specific report are all invaluable for choosing the most appropriate journal.¹⁹ The editorial staff can also help authors if their paper is rejected by explaining the rationale behind the rejection and by providing the reviewers’ comments, which can be used to improve the manuscript.
 9. *Go for it!* Writing a case report takes many more hours of work than most people realize, and there are no guarantees that it will be published. Although the “lowly” case report may not be considered as valuable as other forms of scholarly work for the purposes of promotion,²⁰ we believe that physicians should still “go for it.” Thoughtful descriptions of cases still have a role to play in the advancement of medical knowledge.²¹⁻²⁴ Writing a case report is an excellent learning experience that promotes critical thinking, and it may lead to further scholarly work or spark an unrecognized interest in research. In addition, the case report’s structured format¹⁶⁻¹⁸ forces one to practise concisely written communication.
- The growth of electronic medical journals on the Internet, which are less constrained with respect to space, may provide additional opportunities for the publication of case reports. Other possible venues for sharing the insights related to the case are departmental or hospital rounds as well as regional and national meetings.
- We hope that this article will encourage physicians, especially house staff, to consider writing case reports when faced with the appropriate patient. The tips and suggestions that we have outlined may improve the chances of this process being a more rewarding experience.

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References

1. Thayer WS. Osler, the teacher. In: *Sir William Osler, Bart.* Baltimore: Johns Hopkins Press; 1920. p. 51-2.
2. Huth EJ. *Writing and publishing in medicine.* 3rd ed. Baltimore: Williams & Wilkins; 1999. p. 103-10.
3. American Medical Association. *Graduate medical education directory 1997-1998.* Chicago: The Association; 1997. p. 25-31.
4. Petrusa ER, Weiss GB. Writing case reports: an educationally valuable experience for house officers. *J Med Educ* 1982;57:415-7.
5. Nahum AM. The clinical case report: "Pot boiler" or scientific literature? *Head Neck Surg* 1979;1:291-2.
6. Squires BP. Case reports: What editors want from authors and peer reviewers. *CMAJ* 1989;141:379-80.
7. Beauchamp T, Childress J. *Principles of biomedical ethics.* 4th ed. New York: Oxford University Press; 1994. p. 142-6.
8. Jonsen A, Siegler M, Winslade W. *Clinical ethics.* 3rd ed. New York: McGraw-Hill; 1992. p. 40-3.
9. Hoey J. Patient consent for publication — an apology. *CMAJ* 1998;159:503-4. Available: www.cma.ca/cmaj/vol-159/issue-5/0503.htm.
10. Hoey J. The CMA's Health Information Privacy Code: Does it go too far? *CMAJ* 1998;159:953-4. Available: www.cma.ca/cmaj/vol-159/issue-8/0953.htm.
11. Nylenna M, Riis P. Identification of patients in medical publications: need for informed consent. *BMJ* 1991;302:1182.
12. Slue WE. Unmasking the lone rider. *N Engl J Med* 1989;321:550-1.
13. Bernicker E, Atmar R, Schaffner D, Greenberg S. Unanticipated diagnoses found at autopsy in an urban public teaching hospital. *Am J Med Sci* 1996;311:215-20.
14. Middleton K, Clarke E, Homann S, Naughton B, Neely D, Repasy A, et al. An autopsy-based study of diagnostic errors in geriatric and nongeriatric adult patients. *Arch Intern Med* 1989;149:1809-12.
15. Landefeld C, Chren M, Myers A, Geller R, Robbins S, Goldman L. Diagnostic yield of the autopsy in a university hospital and a community hospital. *N Engl J Med* 1988;318:1249-54.
16. Huth EJ. *Writing and publishing in medicine.* 3rd ed. Baltimore: Williams & Wilkins; 1999. p. 7-11.
17. DeBakey L, DeBakey S. The case report. II. Style and form. *Int J Cardiol* 1984;6:247-54.
18. Rossdale PD, Wade JF, Jeffcott LB. Preparing a case report for publication. *Equine Vet J* 1988;20:7-8.
19. DeBakey L, DeBakey S. The case report. I. Guidelines for preparation. *Int J Cardiol* 1983;4:357-64.
20. Beasley BW, Wright SM, Cofrancesco J, Babbott SF, Thomas PA, Bass EB. Promotion criteria for clinician-educators in the United States and Canada: a survey of promotion committee chairpersons. *JAMA* 1997;278:723-8.
21. Coccia CT, Ausman JI. Is a case report an anecdote? In defense of personal observations in medicine. *Surg Neurol* 1987;28:111-3.
22. Morris BA. The importance of case reports. *CMAJ* 1989;141(9):875-6.
23. Morgan PP. Why case reports? *CMAJ* 1985;133(5):353.
24. Duke M. The well written case report. An asset to the medical literature. *Conn Med* 1974;38(4):169.

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