"In my hands": Part 9: The case for Case Reports

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INTRODUCTION

Evidence based practice (EBP) was developed to assess available scientific evidence and rank it according to the rigour, strength and precision of the research. It aims to provide guidance for clinicians on which to base therapeutic decisions.[1] A number of different hierarchies of evidence have been developed to enable different types of research to be ranked. Systematic reviews, meta analyses and randomized controlled studies (RCTs) usually rank highest because they provide the most reliable evidence of treatment effects. Case reports generally rank low on the scale, just above ideas, editorials and opinions.[2] This is because they are susceptible to bias, have no control group, and cannot be used to establish causal relationships between the intervention and the outcome.[3] However, systematic reviews and RCTs do have a number of limitations, particularly when applied in the evaluation of rapidly developing technologies, therapeutic devices and procedures, or where it is legally or ethically unacceptable to conduct such studies.[3]

No hierarchy of evidence is unanimously accepted.[4] Evans cautions that the use of any of the available measures should be as a guide and not as an inflexible rule.[5] He stressed that different research designs are needed for different clinical scenarios and proposed a different ranking system for studies in the healthcare setting where the main focus is on the effectiveness of the intervention.[5] The strength of such research is evaluated in terms of three components, namely, its effectiveness, appropriateness and feasibility, each scored on a four point scale as excellent, good, fair or poor.[5] He further argues that the benefit of this evaluation in clinical practice is the acknowledgement that many factors can impact on the success of the intervention.[5]

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ACRONYMS

EBP: Evidence based practice
RCTs: Randomized controlled studies

Effectiveness refers to whether the intervention worked as intended, and was appropriate for that particular patient. It also considers the advantages and disadvantages, as well as who will benefit from the intervention. Many believe that systematic reviews and multicentre RCTs provide the best evidence for effectiveness because they represent a range of different populations, settings and circumstances.

Appropriateness relates to the psychosocial aspects of the intervention such as its impact on the person's life, its acceptability and whether the results can be used by others and be generalized as applicable to a wider population (this is known as external validity). It entails considering what health issues are important to patients, and whether they view the outcomes as beneficial.

Feasibility concerns the impact the intervention has on an organization or service provider, the resources required to implement it successfully, whether it will be accepted and used by clinicians, as well as the economic implications to both practitioner and patient.[5]

These three factors also stress that no matter how effective an intervention or treatment may be, if it cannot be appropriately implemented or is unacceptable to patients, then its value is questionable.[5]

Where it is not possible, feasible or ethically acceptable to conduct RCTs, observational studies and case reports may be the most suitable alternative. For example, when measuring infrequent adverse outcomes, or the evaluation of interventions designed to prevent rare events, or in situations where clinicians or patients will not ethically

or legally accept randomization as a means of assigning treatment or not, or where results of RCTs consistently contradict the findings from observational case studies.[5] Thus although case reports have been considered as the "lowest" level of evidence, they are often the "first line" that can lead to further research.[6] This has led to a resurgence in recognising the merits and acceptability of well documented case reports appearing in the literature.

Guidelines on conducting and writing case reports.

1. Why are they useful?

- Case Reports involve genuine clinical scenarios, which makes them more applicable and interesting to colleagues.
- They provide vivid accounts and as such have high readability value and impact.[7]
- They are a means of disseminating knowledge rapidly and succinctly to a widespread related audience.[4]
- They are a unique way of communicating anecdotal observations and to provide brief clinically related findings where experimental evidence is lacking.[8]
- They can be the first pointers in the recognition and description of "new" diseases.[6]
- They can be used to present accounts of rare manifestations of a disease.[6]
- Although they rank far below RCTs on the evidence ladder, they are far less costly and time intensive to conduct. They often shed light on new diseases, patient anomalies, drug effects, and different approaches to treatment that would be impossible to investigate with RCTs for ethical reasons.[8] Note: "The lack of evidence is not the evidence of a lack of effect."[9]
- They educate colleagues about new developments and possible alternative techniques that could be implemented in practice.
- They forewarn others of possible adverse reactions to drugs or materials.
- They alert and inform colleagues of situations where an unanticipated yet informative outcome had occurred during the routine course of treatment. This may help educate and equip practitioners to identify and to handle similar situations in their own patients.[8]
  (You see only what you look for, you recognize only what you know! Dr Merril Sosman)
- They may warn about clinical difficulties or failures encountered with new products.
- Case reports are a good means of evaluation in instances when observational studies reveal consistent differences to findings reported in RCTs.[5]
- They present a more realistic picture of clinical outcomes as opposed to many controlled studies which often report only on the best achievable results.[3]
- They can be used as a discussion forum for ethical dilemmas faced by clinicians. This could prompt further discourse amongst colleagues, ideally leading to unbiased, rational collaborated ideas and solutions.
- They can cover a broad range of topics, and offer a critical opportunity of showing bizarre cases to colleagues.[8]
- They present a full picture of clinical care on a case-by-case basis.[8]
- They serve as a means of continuous education for colleagues.[10]
- New ideas gathered and documented in case reports often generate further research and thus engender an advancement of medical science.[8]
- They cost far less than RCTs to conduct.[5]
- They are a valuable means of educating dental students.[8]

2. Who should write them?

Case reports are based on personal experiences. The "subject material" is encountered during daily practice and as such is easily within the grasp of clinicians. This makes case reports a good starting point for those practitioners wanting to begin research or scientific writing.

3. What are they and what do they entail?

Initially case reports were short, anecdotal communications between colleagues about unique or interesting patients seen in their rooms.[10] Today, reports need to be more structured with a clear description of the case/s, a focused literature review related to the topic, a discussion of how the case may relate to future clinical practice, and should conclude with practical recommendations of suggestions on how to improve the *status quo*.[10] Included should be an initial diagnosis, patient consent, a brief description of the natural disease course and routine treatment if applicable, details of the intervention / drug / therapy / treatment, measure of the outcomes, patient perceptions, remarks on safety and any known risks, and the conclusions of the clinician.[7] In the situation where a series of cases are presented the authors must state whether ALL consecutive patients were treated in the same manner, or why any were excluded, and must report on the outcomes of each case.[7]

4. When should they be written?

The value of case reports lies both in their novelty and / or educational value. Cases that increase the awareness of an unusual condition, describe a rare presentation of a common condition, identify innovative treatment, illustrate the application of a new material, highlight problems with a currently used product, offer advice on improved diagnostic strategies or debate clinical ethical dilemmas, are all constructive and worthwhile contributions to the literature.[11]

They should also be written when a new approach or material has not succeeded. This will alert other colleagues to potential problems, forewarn them before they duplicate the mistakes, and protect patients from costly and potentially harmful procedures.

5. Where are they conducted and published?

The beauty of case reports is that they are based on genuine patients seen in clinical practice. This makes them the ideal avenue for non-academics to follow if they have an interest in sharing knowledge and experiences with others. They are one of the easiest and most rewarding ways to begin writing and publishing for less experienced researchers. They are also an excellent way of fostering collaboration and teamwork as many of the situations involve consultation between private practitioners, manufacturers and academic colleagues – all of whom can be involved in the report.

As a rule, case reports should only be published in peer reviewed journals. However, there has been an emergence of social media groups who share interesting cases with each other. This carries a number of risks for practitioners and the relevant ethical, legal, professional aspects have

been explored in detail in (part 8) the previous paper in this series.

6. How should they be constructed?

Carleton and Webb (2012) suggested that case reports have five sections, and proposed guidelines for each of these as follows:

- A brief Abstract that summarizes the case, its clinical relevance and educational value. It should be concise, and clearly intelligible to colleagues as well as the broader scientific community.[10]
- An Introduction outlines the important clinical aspects and how they relate to current practices. It should also make reference to other key publications on the topic, but is not an extensive literature review.[10]
- The Case is then presented as a chronological description of the patient(s). This includes age and sex, main complaint, initial presentation, co-morbidities, medical, family and social history, examination, diagnostic tests, treatment and materials used (if applicable) and outcomes. Albrecht, Werth and Bigby added that it should include perceptions of the patient(s), and in case series there should also be a follow-up as the results in those who return for recall visits may be a lot different from those who don't.[7] The report should include enough information to justify the intervention and support the conclusion. The treatment should be described in detail with mention of whether it was a once-off, or if it is ongoing, and for how long.[7] The body of the report may contain illustrations, charts, tables or clinical photographs (obtained with the patient's consent and having distinctive features blacked out to ensure anonymity). It should also try to pinpoint some distinguishing patterns that will allow others to recognize similar situations in their practices.
- The Discussion should justify the clinical decisions taken. This can be done by comparing and contrasting with other similar cases, and by making reference to related literature (if any is available). It should focus on one central theme, giving a detailed account of all actions taken, outcomes, and related findings. The authors should stress limitations of the study and offer some "take home lesson". These recommendations may be an augur future treatment modalities or research, and are arguably the most important part of the case report. They should be substantiated by a critical appraisal of the literature, and where possible, try to predict conceivable implications if the findings are generalized and used by others.[10]
- The Conclusion should be a brief, but precise, summary of the central theme, the findings, and the implications for the future. It should be based on the evidence presented as well as the discussion, and must end with a clear message to justify its relevance.[10]

7. What does NOT constitute a case report?

- "Brag books" of before and after treatment photographs.
- Me, myself and I mini-advertisements showing the clinician's special skills, high-tech equipment or state of the art practice and practices.
- Routine treatment procedures given a new lease of life by adding personal commentary.
- Sensational descriptions of bizarre observations.[6]
- Reports that contain misleading elements.[6]
- Proposal of treatment modalities that may do more harm than good if followed by others.[6]
- False alarms which could destroy the credibility of well-tolerated drugs and accepted practices.[7]

8. Drawbacks

The major limitation of case reports is the absence of a control group and the inability to conclusively state that the observed changes are a direct result of the intervention.[3] Kanduluru *et al*. caution clinicians to be mindful of the limitations of using such changes as a basis for future treatment modalities. They argue that other forms of scientific research are evaluated in terms of the levels of controlled evidence available, whether the cause preceded the effect, the clinically relevant pre-trial hypothesis, if the sample size was adequate, whether the study population is properly randomized, if there any different explanations that could work as well or better, whether there is a placebo, and if it was real or sham, and what protection was there against conflict of interest?[2] However, few of these criteria can be applied to any evaluation of case reports. Limited conclusions can be drawn as uncommon side effects may not be seen, thus it is impossible to predict the safety of a procedure or intervention, based on a case report.[7] Be skeptical of those reports which conclude that the treatment is "safe and effective".[7]

Others also warn that case studies have a risk of distorting the treatment effects, making them appear smaller or larger than they actually are.[5] In some situations outcomes are not quantifiable or measureable and rely on the subjective opinions of the clinician and the patient, which are unreliable.[7] A further limitation is the issue of author selection bias and publication bias. In the former, the authors decide what and when to publish, and may report on only a few selected cases. In the latter, journal editors may be less likely to publish reports which do not have interesting data, show dramatic treatment effects, or depict sensational complications.[3,7] In addition, case reports attract fewer citations, making editors reluctant to publish them in favour of controlled scientific studies and meta analyses.[4,7]

CONCLUSION

This paper presents the many merits of case reports, specifically that they are based on real life situations that closely reflect clinical situations, that they can report on rare or infrequent situations and cases where it would not be legally or ethically permissible to conduct a RCT, that the topic material is found in the daily ambit of clinical practices, and the procedure for writing is within the grasp of all clinicians. In the words of Sir William Osler "Physicians should always note and record the unusual".[12] Extrapolated for practitioners, the pertinent points are "Perceive, Publish or Perish!"

PS. In case you need more convincing as to their value, consider that the ground-breaking heart transplant surgery of Professor Christian Barnard in 1967 was first presented to the world as….. a case report![13]

Case Closed!

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