Column: Research methods in Psychiatry

CASE REPORT AND CASE SERIES

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Submitted online: 16/5/2020 Published online: 23/5/2020

ABSTRACT

Case reports/series are the basic types of descriptive epidemiologic studies. The distinction between case reports and case series on the one hand, and case series and descriptive/analytic epidemiologic studies, on the other hand, is not sharp. History of medicine has demonstrated how case reports/series had generated or furthered medical knowledge in many situations. External validity and novelty are the key advantages of these studies. They act as 'screening studies' for deductive epidemiological research. Case reports/series are fraught with biases and lack of strength of evidence to inform clinical decision making. To generate evidence-based information, authors of case reports/series should give attention to the guidelines for maintaining of methodological rigour.
Keywords: case report, case series, descriptive studies, epidemiology

INTRODUCTION

Case reports are the most basic type of descriptive studies that deal with individuals. Case series aggregate individual cases in one report.[1] Case reports/series are uncontrolled (non-comparative). They include a relatively small number of individuals.[2] Distinction between the two is debated—case reports generally do not include more than four cases. In comparison, case series can include more than four cases.[3] The distinction between the case reports and case series is much more than the difference in the number of cases.

CASE REPORT

A case report is a "detailed description of a few patients or clinical cases with an unusual disease or complication, uncommon combinations of diseases, and unusual or misleading semiology, cause or outcome."[4] It can also describe new or innovative approaches to a disease.[5] A good case report may not inform treatment decisions, but it can contribute to new avenues for research. It could propose innovative approaches to diagnosis/treatment or spur the development of hypotheses that can be tested by using other

Please cite this article as: PV Indu, K Vidhukumar. Case report and case series (Column: Research Methods in Psychiatry). Kerala Journal of Psychiatry 2020;33(1): 88-91. doi: 10.30834/KJP.33.1.2020.197

study designs.[6] In 1910, James Herrick reported the case of a 20-year-old patient who suffered from anaemia and manifested 'freakish,' elongated red blood cells, which led to the diagnosis of sickle cell disease.[7] Jordan (1961) reported the case of a 40-year-old lady who developed pulmonary embolism after five weeks of oral contraceptive use, which stimulated other studies that showed a significant association between the use of oral contraceptives and pulmonary embolism.[8] However, some case reports simply enliven drab medical literature.[1]

CASE SERIES

A case series is a "collection of subjects with common characteristics used to describe some clinical, pathophysiological, or operational aspects of a disease, treatment, exposure, or diagnostic procedure."[4] Case series, unlike case reports, is an abstraction of a series of cases collected over a period of time, which lead to hypotheses regarding new diagnoses, interventions, or side effects which could be tested further with methodological rigour.[3,9] A case series of four, previously healthy, people with a sexual preference for the same sex presenting with *Pneumocystis carinii* infection published in 1981 suggested that these patients were suffering from a previously unknown disease, which was subsequently called Acquired Immunodeficiency Syndrome (AIDS).[10] The discovery of the effect of lithium salts on mania was brought out by John FJ Cade (1949) in a case series of ten patients with manic-depressive psychosis.[11]

Case series can be formal or informal (based on whether selected for a specific reason or not), consecutive or non-consecutive, retrospective or prospective, clinic- or population-based and based on exposure or outcome.[2] Case series can be synthesised narratively or quantitatively.[2] Distinguishing quantitative and prospective case series from descriptive and cohort studies can sometimes be challenging. Clearly defined protocol before the onset of the study, statement of inclusion and exclusion criteria and description of loss to follow up are characteristics which differentiate other descriptive/cohort studies from quantitative/prospective case series.[2]

STRUCTURE OF A CASE REPORT/SERIES

In general, all case reports/series should have the following components: abstract, introduction, case presentation, discussion and conclusion. Abstracts are short and should summarise the case, the problem addressed, and the message conveyed. The introduction gives a brief overview of the problem addressed by the case, citing relevant literature. The presentation of the case(s) includes a description of the patients' socio-demographic characteristics, as well as details of history, physical examination, investigations done, treatment given, expected and actual outcomes. The discussion should expand on the introduction focussing on why the case and the problem that it addresses is noteworthy. This should be followed by a review of relevant literature, describing existing theories and research findings relevant to the challenges of the case. The report can end with a conclusion or summary points, with suggestions or recommendations for future

research.[12] To the authors' opinion, case series with quantitative synthesis should be described as other descriptive studies.

ADVANTAGES

A major advantage of the case report is that it can detect novelties; anything that is rare never observed earlier and might be necessary for the medical community. It can lead to the formulation of new hypotheses, which can be further tested with formal, observational or experimental research methods. When experimental research is impossible, as the disorders are rare or prohibited by ethical constraints, less rigorous designs like case reports/series might be the only evidence available. The cost involved is also low compared to formal studies. It can lead to or contribute to a change in clinical practice—change the attitude to, or treatment of, a condition or even lead to the withdrawal of a drug from the market. A letter to the editor published in The *Lancet* in 1961 spurred several case reports and case series, which brought the teratogenic side effect of thalidomide to the attention of the medical community and further led to the withdrawal of the drug. Moreover, clinical medical learning is largely case-based; case histories or vignettes are often presented in textbooks and lectures.[13] In Psychiatry and Psychology, where the objective of the research is to develop an understanding of meaning and intentionality for individual(s), rather than generalisable knowledge, case reporting can be a way of presenting outcomes with idiographic emphasis. These are not case reports per se, but case studies. This approach borders on qualitative research.[14]

LIMITATIONS

Case reports or series do not generate epidemiological quantities like rates, ratios, incidence or prevalence, as there is no denominator involved. Cases are not chosen from representative samples, and there is no control group. Hence, causal associations cannot be inferred from these reports, and the findings cannot be generalised. Overinterpretation or misinterpretation—i.e., a tendency to generalise when there is no justification for it—is called "anecdotal fallacy." Mostly, case reports are retrospective, leading to the occurrence of recall and information (observer) bias. Further, publication bias—due to case reports with positive treatment outcomes being published more frequently than treatment failures—can be a limiting factor.[13] Generally, it has been argued that the treatment approach which had been found useful in a case report might prove otherwise in formal investigation.[5]

METHODOLOGICAL QUALITY OF CASE SERIES AND CASE STUDIES

Although case reports are quite common, their quality is not evenly maintained. The CARE Guidelines is a 13-item checklist, which provides a framework to ensure the completeness and transparency of a published case report. It recommends a timeline—a table or figure—to give the critical dates and times of essential components of the case to show how the key events of the case unfolded. It advocates for patient-reported outcomes and

experiences whenever possible. Informed consent and, wherever necessary, approval of the ethics committee or Institutional Review Board are also included.[15] There are also tools for evaluating the methodological quality of case reports and case series. Most of these tools emphasise the selection of patients, ascertainment of exposure/intervention and outcome, how criteria for causality were ascertained and complete description of the cases.[2]

CURRENT STATUS OF CASE REPORTS/SERIES

Case reports have a low ranking in the hierarchy of evidence. With the emergence of new research methodologies suitable for evidence-based medicine, case reports were side-lined in the second half of the twentieth century. Still, it started regaining popularity by the late 1990s. The *Lancet* introduced a peer-reviewed "Case reports" section in 1995. In 2007, BioMed Central launched the *Journal of Medical Case Reports*—the first international, PubMed-listed medical journal for publishing case reports only. A registry for case reports, Cases Database, founded in 2012, included 11,000 published case reports from 50 medical journals, and it is growing. Innovative modifications like the "storied case report," the "evidence-based case report," the "interactive case report," and the "narrative and evidence-based case report" have been tried without generating a lasting impact. [13]

CONCLUSIONS

Although case reports/series occupy the lowest position in the evidence hierarchy, they provide rich resources for understanding the history of medicine, can facilitate communication between practising clinicians and academic researchers and form an opportunity for young doctors to author an article and prepare for a scientific career.[13] Most importantly, they can generate hypotheses that could be tested further. To circumvent the inherent limitations and to tap the potential opportunities of clinical observation, authors of case reports/series should follow guidelines for quality assurance.

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