The recent history of the clinical case report: a narrative review

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Summary

Clinical case reporting in the form of case reports and case series reports has always been an integral part of medical literature. From the late 1970s the genre appeared to fall from grace and was marginalized in many medical journals. There was controversy as to its value as a research method. From the late 1990s and onwards, there has been an increased demand for and publication of case reports and case series. The various causes for its decline and subsequent return are discussed with an emphasis on the recent historical context.

Background

From personal experience with writing and publishing case reports, we have learned that the chances of finding medical journals that will accept them for publication have varied much over the last decades.[1] There can be several reasons for this. By browsing the literature on case reporting we came across other authors who have described challenges in getting their case report published.[2] This inspired us to take a closer look at the recent history of the case report genre. Our aim of this narrative review is to describe and discuss the changes in some central medical journals' publication policies.

Methods

PubMed and Google Scholar were searched using the terms 'case report(s)', 'case series report(s)', ' case reporting' in combination with 'medical', 'clinical', 'anecdotal', 'review', 'editorial', 'editorial publication policy'. Further references were identified by examining the references found in relevant electronic searches. Some major textbooks on epidemiology and medical research were also consulted.[3,4] We selected relevant articles and book chapters before synthesizing the data.

Defining the clinical case report

A *case* (from the Latin *casus*) can best be understood as a happening, an event or an occurrence. Some consider the patient or person as the case. Strictly speaking, it is the disease or the clinical phenomenon in question that is the case. A *case report* can be defined as 'A detailed description of the experience of a single patient'.[5] Taber's cyclopedic medical dictionary's definition is a bit more elaborate: 'A formal summary of a unique patient and his or her illness, including the presenting signs and symptoms, diagnostic studies, treatment course and outcome'.[6] In the literature, the case report is variously called a *clinical case report*, a *medical case report*, a *single-patient case report,* or a *single case report*. The term *anecdote* is sometimes used in a derogative way to emphasize its rather unscientific status. If more than a single case is reported, it is called a *case series* or *case series report*. These usually consist of 3–10 cases.[7]

The case report is most often a descriptive and naturalistic study as opposed to experimental research. Furthermore, it follows that it is retrospective. It is not part of a planned and designed research project. However, rarely, a case report can be experimental and prospective. In that case it is often called a *case study*. Notable examples of seminal case reports are the discovery of lithium salt in the treatment of psychotic excitement (mania)[8] and the bold first attempt in 1939 at a new surgical procedure, the ligation of a patent ductus arteriosus, on a seven-year-old girl.[9]

Changes in publication policies: a glimpse of the debate

There have been some heated debates regarding the appropriateness of publishing medical case reports. Below, we briefly present parts of a debate occurring in the *British Journal of Psychiatry* in 2003–2005. In 2003, Greg Wilkinson, the editor for the preceding 10 years of the *British Journal of Psychiatry*, commented on his influence on the journal's editorial policy: 'I hastened the demise of the case report, to exclude what I see as psychiatric trivia. I published original research …'.[10] Several readers took to the correspondence section of the journal. First out was D D R. Williams: '…This is a cameo of the polarity that exists between academic[s] … and those clinicians who provide the bulk of the service in the National Health Service… The nomothetic approach takes precedence while the detailed study of an individual patient is marginalized as trivia'.[11] Another proponent is even stronger in his rhetoric appeal: 'Do our patients have loves, hates, hopes, fears, passions, fantasies, beliefs, hobbies, sports? A steady reader of the *Journal* would have no hint that they ever had.'[12] M D Enoch[13] concludes: 'The case history reminds us that the person is not merely a statistic but comprises body, mind and soul and that each must be taken into consideration for complete healing to occur'.

The case report is marginalized

In a study of research designs in clinical literature from general medical journals there was no consistent change in the frequency of case reports involving either single or several cases in the 30-year period from 1946 to 1976.[14] The percentage of case reports was as high as 38. However, the authors defined 'case reports' rather widely as '…data [presented] on 10 or fewer subjects'.[14] Then a steady decline in the number and proportion of case reports in the clinical literature started. A significant reduction of these articles, from 17.4% to 2.4%, took place in the *American Journal of Psychiatry* and *Archives of General Psychiatry* in the 1980s.[15] Concomitantly, there was an increase in the proportion of 'research articles' (defined as 'those which presented original, first-hand data collected in a systematic fashion and which included some clearly delineated methodology') from 50% in 1969–1970 to 82.4% in 1989–1990. The number of research articles had increased by 50%. This change was facilitated by the development of new tools (diagnostic instruments, rating scales, etc.) and new measures for biological parameters.[15]

Editors of several medical and psychiatric journals began to view case reporting as non-scientific in the 1980s and increasingly relegated case reports to the 'Letters to the Editor' section.[16] When evidence-based medicine (EBM) entered the scene, there was even stronger emphasis on large-*n* quantitative studies. The randomized controlled trial ranked high on the evidence hierarchy while case reports and case series were at the lower end. In addition to the prestige of publishing articles with a high ranking on the evidence hierarchy, the journals got more citations, which are the basis for calculating the impact factor. Case reports are, in general, cited less often than reviews, meta-analyses, randomized controlled trials, cohort studies and case-control studies.[17] Publishing case reports, therefore, has the potential to lower a journal's impact factor. Several journals subsequently either banned case reports or applied more stringent criteria (quality, novelty, exceptional interest, brevity, relevance) for accepting them for publication.[18,19]

Another explanation for the low acceptance rate of case reports was the large number of reports being submitted to the hard-copy journals. In other words, a shortage of page space appeared to be a challenge. This is well described by the editors Agha and Rosin[18] in the first issue of *International Journal of Surgery Case Reports*. The *International Journal of Surgery*, a hard-copy journal, had to stem the tide of submissions by

instituting a policy of non-acceptance in 2007. Despite updated instructions to authors, manuscripts of case reports continued to be submitted. The solution seemed to give them a 'dedicated home' in the new sister journal,[18] an online, open access journal launched in 2010.

Case reports are to a large degree written by younger clinicians with limited research experience. This could lead to a low quality of manuscripts and subsequently a lower acceptance rate.

The topic of financial sources is relevant for a discussion of changing trends in publishing. Pincus *et al.*[15] documented a significant growth of research articles in the two major psychiatric journals in the USA reflecting a surge in research activity. In the study period (from 1969 to 1990) there had been an increase in the funding from pharmaceutical companies, although only 2% to 6% of research articles acknowledged financial support from these sources. Another finding was that a significantly lower number of research articles reported no financial support, suggesting that it had become increasingly difficult to publish without some specific source of funding. It seems likely that funding bodies by favouring larger studies facilitated the publication of articles with more advanced designs, thus making case report designs less attractive for authors and journals.

The pendulum swings: the case report returns

In 1995, *The Lancet* introduced a section of peer-reviewed 'Case Reports', with a space limit of one page and 600 words.[19] The journal expected '… younger clinicians, whose daily involvement with patient care is greater than that of their chiefs, to be an especially fertile source of good stories'.[19] In 1997, the *American Journal of Psychiatry,* after a decade not publishing case reports apart from in the 'Letters to the Editor' section, introduced the 'Clinical Case Conference' as a regular feature.[20] This version of the case history had primarily educational value. From the early 1990s onwards several editorials, short reviews and commentaries highlighted the advantages of case reports and case series.[21,22]

What could have caused this renewed interest in case reporting? We have from our review been able to identify seven possible causes.

First, in the 1990s, a fascination with narratives and qualitative research developed in some fields of medicine, especially in general practice and psychiatry. Although most of the published case reports do not follow stringent criteria for qualitative research, they do have some qualitative characteristics. The case presentation is a story, or narrative, of a single individual, sometimes giving an in-depth picture of a patient's life situation. After a couple of decades with a relative scarcity of 'life stories' in the medical literature and an emphasis on quantitative, biomedical research, the medical community seemed ready for both qualitative research and more case reports.

Second, EBM has been met with some scepticism and critique.[23,24] As Greenhalgh[23] stated, ' … the hierarchy of evidence is a hierarchy of clinical epidemiology'. The kind of research that ranks high on this hierarchy is quantitative, large-n studies. As pointed out in Vandenbroucke's[21] often cited editorial in the *Journal of the Royal Society of Medicine*: 'This hierarchy, with the randomized trial on top, holds for a single purpose – the evaluation of the medical interventions with wide applicability in which there is uncertainty about a benefit that is in itself not striking. Case reports and case series have other roles that answer basic needs in medicine'. One of these needs is the progress of science. Case reports are indispensible for its ability to detect novelties, thereby generating new scientific hypotheses.

Third, within psychotherapy research there was in the 1980s and 1990s a resurgence of interest in the intensive study of the individual case.[25] This could maybe, by cross-pollination, have reawakened the medical field's attention to case reporting. One of the methodologies of single-case psychotherapy research, the *N*-of-1 trial, has in recent years been implemented within EBM.[26]

Fourth, in 1998, the *BMJ* launched a new type of article called the 'evidence-based case report'.[27] The intention was to '…[to] help readers develop the increasingly necessary art of using research evidence in practice'.[27] This new kind of case report should not present new findings, but illustrate the diagnostic and therapeutic process. A somewhat different kind of process was presented by Browman in the *Journal of Clinical Oncology.* In presenting a case, he showed how to negotiate good care and treatment in a cancer

patient when the best evidence from EBM was at odds with the patient's preferences.[28] Obviously, various aspects of EBM raise new challenges for clinicians, which again can breed new case reports in want of publication.

Fifth, case reports and case series are popular. Although rarely cited, they have an intuitive appeal, a high readability, and are often read.[18] Clinical practice is case-based, in the sense that medicine is learnt and practiced on a case by case basis. Case reports describe the case-based real world medicine which is about individual patients: at the bedside, in the operation theatre, at morning meetings, etc. Moreover, case reports give nuances and details of practice that one rarely finds in textbooks and lectures.

Sixth, the development of online publishing might have been the factor contributing the most to the revival of case reporting. Since 2007, several online only, open access journals exclusively publishing case reports and case series have been launched. Some of these are independent electronic journals while others are sister journals of established hard-copy journals, e.g. the *BMJ Case Reports* and the aforementioned *International Journal of Surgery Case Reports*.[18] This format removed the limitation of page space. The cost of running most of these journals is covered by a standard article-processing charge levied on articles that are accepted for publication. One of the most important advantages with online publishing is rapid publication. So far, this new brand of medical journals seems quite successful, judging from the quantity of articles published.

Seventh, focus on rare diseases has been advocated. In 2009, the Chief Medical Officer for England dedicated a chapter, 'Rare is common', in his annual report pointing to the challenges of optimizing health care for those with rare diseases.[29] In 2010, the European Commission recommended its members to produce a strategy for these patients.[30] To improve health care for rare disorders that are often diagnosed late or not at all, clinicians' skills must be improved. Stories, i.e. cases, stick to the memory more easily than general knowledge. Having seen or read about a case will make it easier for the clinician to recognize a similar case should she or he encounter it. These arguments for a liberal publishing of case reports might have had an impact on researchers and journals.

Conclusion

The attitudes towards publishing case reports and case series have oscillated with a decline from the late 1970s and then a rise from the late 1990s. We have pointed to several probable causes for the changes. It seems that the decline took place in an era with a strong emphasis on biomedicine and use of epidemiological research tools recommended by the proponents of EBM. The current renewed interest in case reporting can be understood – partly at least – as a counter reaction to this trend. The time had come for 'narrative-based medicine' and a widening scope with a new curiosity for the single individual and qualitative research methods. However, there is a confluence of factors. Undoubtedly, the innovations in electronic publishing have also been very important.

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