

ARTICLE

The case of case reports: a decade of publications by staff at a major university hospital

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Abstract

Rationale: The place of case reports in the medical literature has been much debated in recent years. This study was undertaken to gain knowledge about the publication practice of case reports and case series from a major university hospital.

Method: We decided to conduct a retrospective, bibliographic, descriptive study of published case reports and case series from a university hospital in a period of 10 years. The following variables were studied: number of reports, authorship characteristics, collaboration practices, titles, medical specialities represented, educational *versus* non-educational purpose and number of citations. The data were extracted from the national research information system database in Norway.

Results: 2.2% of all the publications were case reports. Multiple authorship was common. Male authors outnumbered female authors. Collaboration across hospitals and nations occurred for one third of the articles. 43% of the titles did not contain information that identified them as case reports or case series. The most frequently represented specialties were neurology, rheumatology, plastic surgery and medical genetics. Nine out of 10 articles were non-educational. A third of the articles had not been cited.

Conclusions: The case report seems to be a minor, although viable, genre. The proportion of case reports and case series was low. A plausible hypothesis could be that clinician-researchers at the study hospital prioritized controlled clinical and paraclinical/laboratory studies that rank higher on the evidence hierarchy. Since case reports document the presentations of individual patients and their treatment, a declining interest in their publication has a significant implication for person-centered healthcare education and training.

Keywords

Authorship, bibliometric case reports, citations, collaboration practices, medical specialities, person-centered healthcare, titles

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Introduction

Medical writing has been the object of interest and study among medical researchers and scholars of writing studies, literature and linguistics [1-5]. Among the various medical genres, the clinical case report is probably the oldest [6]. Egyptian papyri dating from the 16th-17th Dynasty, that is, 1650-1550 B.C., contain case reports with an emphasis on treatment [7]. These were typical cases, not individual ones, instructing on diagnosis and treatment. The linguist Dwight Atkinson found single cases of disease to be the most common article type in the *Edinburgh Medical Journal* in the 18th Century [4]. In a study of publications from three general medical journals - the *Journal of the American Medical Association*, the

Lancet and the *New England Journal of Medicine* - in the period from 1946-1976, case reports and case series comprised 38% of the articles [8]. The authors defined "case report" as including 10 or fewer patients. Thirteen percent were single case reports. There was no significant decline in this period. The reduction, both in number and proportion, of case reports and case series came later. In the period from 1971 to 1991 there was a reduction from 30% to 4% case reports in the same three journals [9]. In the 1980s, the proportion of the articles fell from 17.4% to 2.4% in the *American Journal of Psychiatry* and the *Archives of General Psychiatry* [10].

The percentage of case reports and case series declined from 19% in 1991 to 1% in 2001 in the journal *Obstetrics & Gynecology* [11]. The trend towards decreasing number of case reports was not ubiquitous,

however. Some journals kept the number of case reports at fairly steady levels, 34-45% and 13% respectively in two studies, from around 1980 to the late 1990s [1,12]. Anyhow, the case report genre plays a minor role in most medical journals today [13,14].

Other areas of interest concerning case reporting have been medical specialty, authorship, collaboration practices, citations, titles and 'instruction to authors' [3,15-18]. The source of data for these studies has usually been limited to one or a few medical journals and the results may thus reflect editorial policies or other unique aspects of these specific journals. We do not know of any previous studies that have focused on the output of case reports from a university hospital. Knowledge about various quantitative characteristics of case reports might inform us about developments and trends in medical practice and research, including editorial policies in general and their significance for person-centered healthcare education and training.

The objective of this study was to explore the publication practice among professionals at a university hospital with regard to case reports and case series, focusing on the following variables: 1) The production of case reports and case series, 2) Issues related to authorship (number of authors per article, gender distribution, gender of first and last author), 3) Collaborative practices (local, national, international), 4) Titles, 5) Medical specialities represented, 6), Educational or non-educational purpose and 7) The number of citations.

Methods

We decided to conduct a retrospective, bibliographic, descriptive study of published case reports and case series from the University Hospital of North Norway which we subsequently term *the study hospital*. We applied *Taber's Cyclopedic Medical Dictionary's* definition of case report, "A formal study of a unique patient and his or her illness, presenting signs and symptoms, diagnostic studies, treatment course and outcome" [19], although with the modification that we allowed for 2 cases in the report. Articles based on hypothetical or simulated cases or case series were excluded [20-22]. A case series or case series report was operationalised as 3-12 cases in this study. Inclusion criteria were: All case report articles, case series articles and case reports-abstracts published in peer-reviewed medical journals, national or international and registered in the Current Research Information System in Norway (abbreviated CRISStin) for the 10-year period 2004-2013 [23]. At least one of the authors had to be employed at the University Hospital of North Norway.

All the articles published in scientific journals by employees of the hospital during the study period were screened for research design. Searching the database allowed for reviewing every article in a stepwise manner. The first step was inspecting the particular article for authorship, title and journal reference. Step two allowed access to the abstract and full text article. Thus, we could

determine research design for all articles. This procedure made for a full catch of all case reports and case series in the CRISStin database for the 10-year period. The number in the database deviates somewhat from the total number of articles produced by the hospital researchers. The database stores only articles that are actively reported by those who have written them. There will be some articles missing, especially in the early years of the period as the current incentives for having articles in the database (e.g., the possibility of receiving financial support, being privileged sabbaticals for those also having university affiliations, *etc.*) were not implemented at that time. Therefore, our material refers only to the corpus of articles found in the database and not the complete number of articles published in the period. The discrepancy between the real and the assembled number is unknown, but we suggest that it is likely to be quite low.

Following identification of all articles, the following data were extracted: (a) The number of case reports and case series, respectively; (b) the number of male and female authors; (c) the gender of the first and last author; (d) the number of articles with local, national and international authorship; (e) whether the title was identifiable or not; (f) the article's primary medical specialty; (g) whether the article had an educational or non-educational aim and (h) the number of citations. The first author is usually the one who has contributed the most to the work. The last author position is usually reserved for the senior author and/or supervisor.

Definitions

The authorship was characterized as *local* if all the authors worked at the study hospital. *National authorship* required authors at two or more institutions in different locations in Norway, with at least one author from the study hospital. If at least one of the authors was working in another country at the time of writing the article, we defined it as *international authorship*. These categories are essentially identical to those of Salager-Meyer and co-workers [3].

A title was labelled *identifiable* if it unequivocally identified an article as a case report or case series, as seen in these two titles:

Progressive multifocal leucoencephalopathy in an immunocompetent patient with favourable outcome. A case report.

Imported case of visceral leishmaniasis presenting in a Norwegian patient treated with methotrexate and etanercept for psoriasis arthritis.

Most titles in the CRISStin database other than case reports were relatively easily excluded when the title contained information on study design, for example, *Chemoradiotherapy of anal carcinoma: Survival and recurrence in an unselected national cohort*. A national cohort is obviously not a case report. Ambiguous article titles had to be meticulously looked into, either by

studying the abstract or the full text version. These are three examples of titles that we discovered to be case reports:

Focal myositis – Aneurogenic phenomenon?

A Little bleed

Arctic environment triggers migraine attacks.

The ambiguous titles were labeled *non-identifiable* as the title itself was not conclusive as to the articles' design or type of study.

Specialty areas for the case reports were identified by the hospital department affiliation (e.g., Gastrointestinal Surgery, Neurology) of the authors. In the case of authors from different departments, we determined specialty according to the first author's specialty affiliation and/or which primary organ system that was described in the text.

Some medical journals, for example, the *New England Journal of Medicine*, the *Lancet*, the *Journal of the Norwegian Medical Association*, have devoted space for regular case reports with an educational aim. These are often supplemented with a commentary by one or several senior specialists. We labelled reports in this category *educational case reports* so as to differentiate them from articles with an ambition of presenting new findings or otherwise further medical knowledge. The last mentioned type was labeled *non-educational*. The number of *citations* for all articles were found by searching the Google Scholar database.

Results

The study material

In total, 2313 published articles from the study hospital were found in the CRISTin database for the 10-year period. Fifty-one were case reports and series (2.2 % of the total number). (Table 1). Forty-one of these were case reports (80%) and 10 were case series (20%).

Table 1 Types of case reports included

Article type	No (%)
Case reports	41 (80)
With 1 case	36 (70)
With 2 cases	5 (10)
Case series	10 (20)

The total number of cases in the 10 case series was 55, giving a mean of 5.5, median 4, mode 3, range 3-12.

Authorship

The total number of authors of the 51 articles was 226. The average number of authors per article was 4.4, with a range of 1-24. If we omit the one outlier article with 24 authors, the mean would be 4.0. The distribution was skewed to the right, both mode and median values were 3. The gender distribution is presented in Table 2.

Table 2 Gender and authorship position of case reports

Gender	All authors No. (%)	First author No. (%)	Last author No. (%)
Women	67 (30)	13 (25)	9 (18)
Men	159 (70)	38 (75)	42 (82)
Total	226 (100)	51 (100)	51 (100)

Table 3 Collaboration practices in case reports

Level	No (%)
Local	33 (65)
National	11 (22)
International	7 (14)
Total	51 (101)*

*Percentage higher than 100 due to rounding errors on individual percentages in the column above.

Article titles

Twenty-nine articles (57%) contained sufficient information to be identified as case reports leaving 22 (43%) without identifier as to type of study design.

Medical specialty

Table 4 Contribution of case reports from the different medical specialties

Medical specialty	No. of reports (%)
Neurology	9 (17.6)
Rheumatology	8 (15.7)
Plastic surgery	5 (9.8)
Medical genetics	5 (9.8)
Anesthesia / emergency medicine	4 (7.8)
Hematology	3 (5.9)
Infectious medicine	2 (3.9)
Clinical pharmacology	2 (3.9)
Pathology	2 (3.9)
Psychiatry	2 (3.9)
Abdominal surgery	2 (3.9)
Physiotherapy	1 (2.0)
Pediatrics	1 (2.0)
Rehabilitation & physical medicine	1 (2.0)
Medical biochemistry	1 (2.0)
Urological surgery	1 (2.0)
Cardiothoracic surgery	1 (2.0)
Oncology	1 (2.0)

Forty-one reports/series (80.4%) were clinical and 10 (19.6%) were paraclinical (pathology, clinical pharmacology, medical biochemistry, medical genetics). Nine (18% of the total number) of the clinical reports/series were surgical, whereas 32 (63%) were non-surgical.

Educational versus non-educational

Six case reports (12%) were educational, 45 (88%) were non-educational.

Citations

Table 5 Number of citations per case report

No. of citations	No. of articles (%)
0	17 (33.3)
1	11 (21.6)
2	5 (9.8)
3	2 (3.9)
4	3 (5.9)
5	3 (5.9)
6	1 (2.0)
8	1 (2.0)
10	2 (3.9)
11	1 (2.0)
12	1 (2.0)
15	2 (3.9)
18	1 (2.0)
45	1 (2.0)

One third of the articles had not been cited. The mean number of citations per article was 4.0. The median was 1.

Discussion

Case reports

The percentage of case reports from our hospital (2.2%) is yet another indication that the case report has lost its central position in the medical literature [13,14]. As already mentioned, we do not have data from other hospitals for comparison. Therefore, we abstain from passing any judgement as to whether the percentage should be higher or lower. However, we find it interesting that 13% of all articles in the *Australian and New Zealand Journal of Psychiatry* in the 33-year period 1976-1999 were case reports [1]. The authors, after having discussed the advantages and shortcomings of the case report, suggested that the genre should still be retained as full articles in the journal, "... but perhaps

account for less than 13%." Furthermore, as our study was descriptive with a relatively short time span, a low number of reports and some uncertainty about the number of missing reports, we cannot identify any clear temporal trend.

Authorship

The mean number of authors per article was 4.4, the median was 3. This is in line with previous research on case reports [3,24-26]. This is somewhat lower than for research articles with more advanced methodologies [9,26,27]. Only one case report in our corpus had a single author. The trend the last decades has been an increasing number of authors in medical articles in general [9,26,27]. This is partly explained by medicine having become more complex and dependent on team work [9,26], although the phenomenon of honorary or gift authorship might explain some cases of multiple authorships [28,29]. Our study was not designed to reveal whether honorary authorships were prevalent, but we admit being surprised when finding one article with 24 authors. The rhetorical question: "Does it take a village to write a case report?" by Gady Har-El in *Otolaryngologia - Head and Neck Surgery*, seems apposite [29].

As concerns gender distribution, there was a male dominance with a male to female ratio of 7:3 for all authors and somewhat larger imbalances for the first and last author (Table 2). We take this as a corroboration of the findings of other investigators [30,31]. Gender authorship appears to be a valid surrogate marker of the gender imbalance of medical publishing [30]. Among doctors and psychologists (who do most of the research at the study hospital), there were in 2005 59% males and 41% females [32]. As the ratio between the sexes has been equalizing in the years following 2005, we feel justified in assuming that male doctors are somewhat more given to writing case reports than female doctors at the study hospital.

Collaboration practices

During the last decades, a growth in collaboration practices has taken place [33]. A study from Taiwan investigated the country's clinical medical articles indexed in the Institute for Scientific Information (ISI) database (ISI Essential Science Indicators) from the period 1990-2004 [34]. A total of 13.6% of the clinical medical articles were produced with an international collaboration. A Malaysian study found local collaboration accounting for 60.3% and international collaboration for 39.7% of clinical medical articles [35]. Salager-Meyer and co-workers investigated exclusively case reports in their diachronic study of the *British Medical Journal* and *BMJ Case Reports* [3]. For the year 2009, they found that 68.3% of case reports were written by authors from the same city (local collaboration), 26.7% had national authorship while only 1.6% were the result of international collaboration. In our hospital-based study the corresponding numbers were 65%, 22% and

14%. Both Salager-Meier's study and ours had lower rates for international collaboration than the two Asian studies referred to above. This should not come as a surprise as case reports usually require less resources and co-operation than larger and more elaborately designed studies. What really was surprising, however, was the number of single-authored case reports. All the 60 case reports published in the 1840-1850 corpus were single-authored, while this was the case with only 2, that is, 3.3%, in 2009 [3]. In our study, only one case report (2%) was written by a single author.

Article titles

Almost half of the titles (43%) did not suggest that the articles were case reports or case series. This was surprising considering the increasing emphasis on the function of titles as identifiers in bibliographic information retrieval systems [17]. The *Journal of Medical Case Reports* instructs authors to include a study design specifier, either "a case report" or "a case series", in the title [36]. The journal suggests this format for the title: *A presenting with B in C: a case report*. This is an example: *Clinical picture and treatment implication in a child with Capgras syndrome: a case report* [37].

Medical specialty

According to the relative "size" (i.e., clinical importance in terms of number of patients treated, number of specialists, etc.) of the various specialties, some appear to be over-represented (neurology, rheumatology, plastic surgery), some under-represented (psychiatry, oncology) and some missing (e.g., gastroenterology, cardiology, ophthalmology, otorhinolaryngology). The regular column named *Lancet Case Report* has been discussed twice in the *Lancet*. In 2003, three neurologists reviewed all the journal's case reports from 1996 to 2002 [38]. They found that the neurological organ system was disproportionately often presented (29%). It ranked high above the second one, which was gastroenterology with 15%.

At the lower end were psychiatry (2%) and ophthalmology (1%). Seven years later, neurology was still the dominant organ system represented in case report publications, but not to the same degree [15]. For the period 2003-2008, neurology accounted for 15% of case reports. Coles *et al.* suggested the dominance of neurology could be explained by "... the trepidation and interest that neurological syndromes generate among physicians" [38]. A review of the first 100 cases in the *Journal of Medical Case Reports*, a journal established in 2007, found general surgery and general medicine representing 11 each, followed by oncology (7), orthopaedics (7) and ophthalmology with 6 reports [39]. Only one report was neurological.

It seems wise not to put too much emphasis on the various proportions of medical disciplines being reported. A published case report is contingent on many factors: rare occurrence; observant and trained clinicians;

opportunity, willingness and time to collaborate with colleagues in order to produce a draft for an article; etc. [14].

From the study hospital we assume there were no obsolete reports. The high number of reports from neurology, rheumatology and plastic surgery can probably be ascribed to a few enthusiastic and very skilled clinicians in these specialties. It may also reflect different attitudes in the various specialties to the acceptance of writing and communicating patient cases. Furthermore, we might assume there have been some reportable cases that have not materialized as case reports. It is a well-known fact that many busy clinicians have little time for academic work.

Educational or non-educational purpose

A study of the characteristics of case reports published in a Danish general medical journal, *Ugeskrift for Læger* (*Weekly journal for doctors*), found that 124 of 140 reports (89%) had an educational purpose [24]. The main audience of this journal is Danish doctors. It is written in Danish, but articles are supplemented with a brief English summary. The authors assumed that the non-educational articles (e.g., discoveries of new associations) probably would be published in high-impact international journals, leaving the educational ones aimed for Danish readers for publication in the national Danish-language journal (*Ugeskrift for Læger*).

In our study, the proportions of educational and non-educational reports were the reverse. A plausible interpretation could be that clinicians at the study hospital find it more imperative and possibly more rewarding to share non-educational case reports with the medical community.

Citations

Citations of articles registered in the Web of Science allow for calculation of the impact factor of journals and articles. Few citations result in a low impact factor. Although the frequency of citations as a measure of quality and impact is debatable, it is essential in determining the prestige of health sciences journals [16,40]. Among the various study designs in health sciences, the case report seems to have a negligible citation impact [16]. Patsopoulos *et al.* documented a median of 1 citation count per article within the first two years of publication of case reports. Furthermore, less than 1% received more than 10 citations within the same time span. This contrasts with meta-analyses, the highest-ranking study design, for which 32.4% (in 1991) and 43.6% (in 2001) received more than 10 citations.

One third of the articles in our material were not cited. The average number of citations per article was 4.0. As the distribution was skewed, a more appropriate measure of the central tendency is the median, which was 1. Seven articles (14%) received more than 10 citations. A head on comparison with the study of Patsopoulos *et al.* is inappropriate considering the different

methodologies. Some of the articles in our material had a 10-year period to accumulate citations while the basis for indexing in the Web of Science is only the first two years after publication. Furthermore, there appears to be a trend in recent years towards higher citation rates that can partly be explained by the increasing number of journals worldwide [16].

A strength of this study was its innovativeness in which a university hospital was chosen as study object. We have not found any similar studies focusing on the output of cases from this type of institution. The quantitative data from this study could represent a benchmark for similar bibliometric studies from other hospitals, medical centers or health trusts. There are, however, several limitations to this study. The number of case-articles was relatively modest despite reviewing a 10-year period of publications. We had no good comparison studies, that is, similar studies from other hospitals or health trusts. Our source of articles, the CRIS database [23], might not include all the published articles from the study hospital and some reports or case series might therefore be missing. Comparing our findings to the publication policies of individual journals is far from optimal. Still we feel this comparison could be justified as it helped us make some tentative interpretations.

Conclusions

Case reports and case series cannot be planned in the same way as experimental, elaborately designed research can. The production of a case report is thus contingent on a chance occurrence as well as diligent, observant and responsible clinicians undertaking the cumbersome work of producing the case report. In addition there has to be a health science journal willing to review and publish the report. Therefore, the total number of case reports from our study hospital does not allow for any clear conclusions. The proportion of 2.2% among all study designs could be a reflection of the genre's presently rather low status in medical literature. Multiple authorship was common and male authors outnumbered female authors. Collaboration across hospitals and nations occurred for one third of the articles, a significant number considering the lesser need for collaboration of this genre compared with other study designs. A relatively large proportion of titles did not inform adequately about the articles being case reports or case series. Of the medical specialties, neurology, rheumatology, plastic surgery and medical genetics stood out as the most productive, with more than half of the articles. Almost nine out of ten reports were non-educational. The uncitedness rate was close to one third.

Considering that only one in fifty articles was a case report or case series, a plausible hypothesis is that researchers at the study hospital prioritize other kinds of clinical and paraclinical/laboratory research, that is, controlled studies with research designs with a higher ranking on the evidence hierarchy. Since case reports

document the presentation of individual patients and their treatment, a declining interest in their publication has a significant implication for person-centered healthcare education and training.

Conflicts of Interest

The authors declare no conflicts of interest.

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