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ORIGINAL ARTICLE

Analysis of the evolution of the publication of case reports in endocrinology journals $\stackrel{\mbox{\tiny $\%$}}{\mbox{}}$



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KEYWORDS

Case report; Journal; Endocrinology; Publication; Clinical investigator

Abstract

Introduction: A case report is a scientific article describing one or more patients with unusual clinical presentations. In recent years, the number of case reports in publications has decreased. In this study, we analyze the publication of case reports in journals of Endocrinology during the years 2010, 2015 and 2019.

Materials and methods: The Pubmed web was browsed for clinical journals of Endocrinology, those published in English and/or Spanish being selected, and the relevant variables analyzed. *Results:* Of 84 analyzed journals, 51 accepted cases for publication, 29 did not, and 4 did so only in exceptional cases. In 2010, 11,754 articles were published, of which 709 were clinical cases (6.9% of the total); in 2015, a total of 14,594 articles of which 655 were clinical cases (5.8% of the total); and in 2019 a total of 14,110 articles, of which 472 were clinical cases (4.6% of the total). In journals demanding payment for the publishing of clinical cases, case reports represented 9% of all articles, and in free journals, 3% (P < 0.05).

Conclusion: There has been a decline in publication of case reports in journals of Endocrinology in recent years, both in absolute and relative terms. Even though the cases described by these reports are, by definition, exceptional, the decline of their publication implies a significant loss of scientific information and clinical knowledge regarding certain pathologies.

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PALABRAS CLAVE

Caso clínico; Revista; Endocrinología; Publicación; Investigador clínico

Análisis de la evolución en la publicación de casos clínicos en revistas de endocrinología

Resumen

Introducción: El caso clínico es un artículo científico que describe uno o varios pacientes con presentaciones clínicas inusuales. En los últimos años, se ha descrito una disminución en la publicación de casos clínicos. En este trabajo analizamos la casuística de publicación de casos clínicos en revistas de endocrinología durante los años 2010, 2015 y 2019.

Material y métodos: Se realizó una búsqueda en Pubmed, y se seleccionaron revistas clínicas de endocrinología, de lenguaje inglés y/o castellano, analizando variables relevantes.

Resultados: De 84 revistas analizadas, 51 admitían casos para publicación, 29 no lo hacían y 4 excepcionalmente. En 2010 se publicaron 11.754 artículos, de los cuales 709 eran casos clínicos (6,9% del total); en 2015, un total de 14.594 artículos, de los cuales 655 eran casos clínicos (5,8% del total); y en 2019 un total de 14.110 artículos, de los que 472 eran casos clínicos (4,6% del total). En las revistas que exigían pago por publicar los casos clínicos representaban el 9% de los artículos, y en las revistas con gratuidad, estos eran el 3% (p < 0,05).

Conclusiones: Existe un descenso en la publicación de casos clínicos en las revistas de endocrinología en los últimos años, tanto en cifras absolutas como en el porcentaje respecto a la totalidad de los artículos publicados. Pese a que un caso clínico aislado solo es relevante si es excepcional, la disminución de publicación de casos clínicos puede suponer una pérdida de información científica y de conocimiento clínico de determinadas enfermedades.

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Introduction

Historically, the publication of case reports began as a rudimentary means of research and contribution to scientific evidence, at a time when science was less developed and conditions or disease variants that are now known and categorised were then exceptional and surprising findings. Observations based on one or more patients were an accepted, established and common form of scientific publication. Nowadays, their validity and perception are less highly regarded among the scientific community, and they have been displaced by higher-quality research articles such as clinical trials (preferably randomised), meta-analyses or systematic reviews, to name but a few.

The International Epidemiological Association defines case reports as "detailed descriptions of a few patients or clinical cases with an unusual disease or complication, uncommon combinations of diseases, an unusual or misleading semiology, cause or outcome''.¹ Among the advantages offered by scientific articles of this type are that they enable new diseases, atypical presentations of common diseases or the beneficial or adverse effect of a treatment to be showcased, and may lead to hypotheses and serve as a basis for further research on the subject. It was thanks to scientific articles of this type that the discoveries of penicillin, mitochondrial diabetes, acquired immunodeficiency syndrome (AIDS), cystic fibrosis, neurofibromatosis, Edwards' syndrome and the first heart transplant, among others, were made known.²⁻⁴ Another of their advantages is their cost, which may be non-existent or negligible as no economic resources are required for descriptions, as well as the short turnaround time from observing the event to preparing the article for publication, which can be of help in certain alarming emergent situations. $^{\rm 5}$

Their limitations include not being representative of either the general population or a specific population with a particular condition, as by definition case reports involve atypical presentations or characteristics. Another reason why extrapolation from them may be difficult is the low prevalence of the disease or scenario in the general population. Because of this low prevalence, there are few case reports deemed suitable for publication. Moreover, being observational in nature, causality cannot be inferred. These biases are one of their main disadvantages, as they can be highly dependent on personal experience, in addition to the fact that motivation to publish positive atypical data is more common than motivation to publish negative atypical data. It should also be noted that it is at times impossible to guarantee the patient full confidentiality.^{1,5}

Although case reports are only of relevance when describing truly exceptional diseases or presentations, and/or they can have added value when a case series is combined with a literature review on the subject, it should be borne in mind that the scientific evidence they provide is not robust. Case reports often act as a starting point for scientific research for students, junior doctors, research fellows, etc. Nevertheless, it is not unusual for authors to encounter difficulties publishing case reports submitted to scientific journals. Some studies have noted an increase in the publication of case reports: specifically, in 2010, 45% more publications of this type were indexed in the PubMed and Embase databases.¹ Even so, in recent years there have been few articles that have investigated the evolution of the number of publications and whether this increase is an isolated event or a trend.

The main objective of this study is to describe the reality in recent years with regard to the publication of case reports in endocrinology clinical scientific journals, and the ratio of case reports to the total number of publications in these journals, as well as to describe the trend in the publication of case reports in these journals over the period studied. A secondary objective is to find out whether there is a link between paid and free models for the publication of articles in scientific journals and the publication of case report-type articles.

Material and methods

Firstly, а search was conducted in PubMed (https://pubmed.ncbi.nlm.nih.gov/) to identify endocrinology clinical journals indexed in Medline. This search was performed in the National Library of Medicine database, using the key word Endocrinology, and subsequently filtering for journals indexed in Medline only. Finally, those in English and/or Spanish were selected. The following variables were collected for each journal studied: whether the journal accepts case report-type articles; whether it charges a fee to publish: the total number of articles published in the years 2010, 2015 and 2019; and the number of case reports published in the years 2010, 2015 and 2019.

Information on variables relating to acceptance of case report-type articles and publication fees was obtained from that available on each journal's website on the dates the study was conducted, between January and June 2020, with publication considered to be ''free of charge'' for those journals that specified that payment was optional, monetary expenditure therefore not being obligatory. With regard to whether a journal accepts case report-type articles, based on the information available on its website, those journals that expressly specified them in the list of article types accepted or those with a ''short article'' section were considered to do so. In contrast, journals were considered not to accept case reports if they specified this or if they required an express invitation.

Journals were categorised as "review journals" if, according to their website, they only considered review articles, and not original articles, for publication; in contrast, those that included original articles among the types published were considered "non-review journals". The number of cases published in each journal during the years 2019, 2015 and 2010 was obtained from PubMed, as was the number of case report-type articles published in each journal in the same years, using the "case report" filter.

The impact factor of each journal for each of the years analysed was obtained from *Journal Citation Reports*. For some journals/years, the impact factor could not be obtained from this source and was found using a web search.

For each year, a descriptive statistical analysis was conducted of the articles published in each of the journals studied, of the case reports published and of the percentage of case reports published out of the total number of articles. Descriptive statistics (median and interquartile range [IQR]) were also used to analyse the number of journals that accepted the submission of case reports for publication and those charging a fee to publish. The Wilcoxon signed-rank test was used to analyse whether differences existed between journals charging a fee to publish and those not doing so in terms of the number of case reports published by each journal and their percentage of the total articles published.

For each year, and excluding review journals, the Pearson correlation coefficient between the number of case reports published in each journal and its impact factor for that year was calculated.

The R version 4.0.2 statistics software in the RStudio integrated development environment was used for the statistical analysis.

Results

A total of 84 journals were analysed. Appendix B - additional material shows for each of them whether they accept the submission of case reports for publications, whether they charge a fee for publication, whether they are considered to be review journals, the total number of articles and case reports published in the years 2010, 2015 and 2019, as well as their respective impact factors for each year.

Twelve of the total were review journals. Regarding whether case reports were considered for publication, 51 (60.7%) did accept them, 29 (34.5%) did not and 4 (4.7%) did so only in exceptional cases. Regarding publication fees, 14 journals (16.7%) charged a fee, 60 (71.4%) did not and for 10 (11.9%), this information could not be obtained from the journals' websites. After excluding review journals, 68.1% accepted case reports, 26.4% did not and 5.5% did so only in exceptional cases; 19.4% of journals charged a fee, 75% did not and for 5.6%, this information could not be obtained from the journals' websites.

Table 1 shows, for each year, the total, mean, standard deviation, median and interquartile range of the articles published, case reports published and percentage of case reports out of the total articles published.

Upon analysing the journals that had published at least one case report in the years studied and whose websites specified whether a fee was charged for publication, it was found that in the 12 journals that charged a publication fee, a total of 769 case reports had been published, with a median (IQR) of 36 (73) cases per journal, while in the 44 that did not charge such a fee, 1030 case reports had been published, with a median (IQR) of 10 (32) case reports per journal, a difference that was statistically significant (p = 0.022).

With regard to the percentage of case reports out of the total articles published, in the journals that charged a fee, the percentage of case reports published out of the total number of articles was 9% (12), while in those with no fee, the percentage was 3% (5), a difference that was statistically significant (p = 0.032). Fig. 1 shows this association. This difference remained statistically significant (p = 0.046) after excluding review journals.

Having excluded journals dedicated to publishing reviews from the analysis, a weak negative correlation between the proportion of case reports published in each journal and its impact factor for the same year was found for the years 2010 and 2015 (r = -0.35, p = 0.008 for 2010; r = -0.35, p = 0.004for 2015), but not for 2019.

	2010	2015	2019
Articles published			
Total	11,754	14,594	14,110
Mean (SD)	140 (153)	174 (155)	168 (129)
Median (IQR)	89 (130)	128 (156)	147 (166)
Case reports published			
Total	709	655	472
Mean (SD)	9 (16.9)	7.8 (13.6)	5.6 (9.5)
Median (IQR)	1 (12.5)	1 (10.0)	1 (6.25)
Case reports as a percenta	ge of total articles		
Mean (SD)	6.9 (11.6)	5.8 (10.8)	4.6 (9.7)
Median (IQR)	1.1 (7.4)	0 (5.8)	0 (4.8)
Percentage of case reports	excluding review journals		
Mean (SD)	7.9 (12.3)	6.7 (11.4)	5.2 (10.3)
Median (IQR)	1.5 (8.5)	1.3 (6.2)	1.2 (6.2)

Table 1 Articles published, case reports, and case reports as a percentage of total articles published in the journals studied for the years 2010, 2015 and 2019.

IQR: interquartile range, SD: standard deviation.

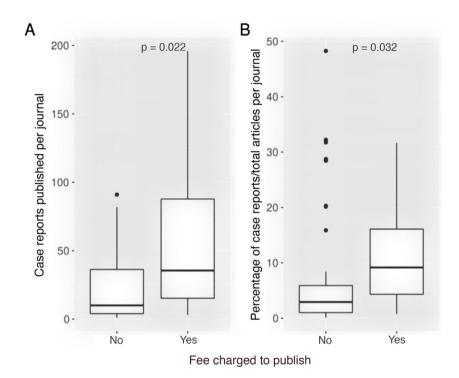


Figure 1 (A) Case reports published per journal by fee-charging status. (B) Case reports as a percentage of total articles per journal by fee-charging status.

Discussion

The dissemination of an isolated case report is only of relevance if it is truly exceptional and/or in rare diseases or new diseases about which little is known. In contrast, it carries added value if it is accompanied by the analysis of a small series, although in rare diseases or presentations, where it is unlikely that a single centre would treat two cases, it can be difficult to describe case series in the absence of registries or isolated cases previously published in the literature. A fall in the publication of case report-type articles has been reported in recent years,⁶ although there are few studies investigating this in detail. In our study, a marked reduction of almost one third was found in both the total publication of case reports in endocrinology journals (709 case reports published in 2010 versus 472 in 2019) and as a percentage of the total articles published (from 6.9% to 4.6%). Our data are consistent with the studies conducted on journals of other medical specialties. In high-impact otorhinolaryngology journals, Eldemeyer et al. found an increase in the number of articles published but only a marginal increase in case reports, with a reduction in the percentage of case reports from 15% in 2010 to 8.82% in 2014 and 8.5% in 2015.⁷ Another similar study conducted on the 10 principal paediatrics journals found a reduction in case reports from 30% 40 years ago, to 15% 20 years ago and 4% in 2016.⁸

The data obtained in our study are along the same lines, both in the drastic reduction in published case reports, and in proportion to the total number of articles. In spite of the scarcity of studies investigating the current trend in case report publication and the differences that there may be in the type of articles generated as scientific evidence for specialties as disparate as otorhinolaryngology, paediatrics and endocrinology and nutrition, the data all point to a reduction in case report publication in the scientific literature of today, along with the loss of knowledge that the omission of exceptional presentations, rare correlations between diseases, serious but extremely rare adverse effects, etc. may entail.

Moreover, it is striking that, based on the results of our study, fee-charging journals publish more case reports than journals in which publication is free of charge, both in absolute terms and in proportion to the total number of articles published (9% of total articles published in fee-charging journals versus 3% of all articles in journals with free publication, p < 0.05). Moreover, according to our results, journals with a greater impact factor tend to publish fewer case reports, although the correlation is very weak.

The limitations of this study are primarily its descriptive, cross-sectional methodology, as well as the fact that only endocrinology journals found in our PubMed search could be included and therefore analysed. A further limitation is that only case reports catalogued as such were included, while others indexed in such sections as ''letters to the editor'', ''images in medicine'' or ''virtual case reports'' may have been missed. Although the relationship between the journals' impact factors and whether they accept case reports was analysed, the true significance of each case report can only be inferred by analysing the number of citations it receives; such an analysis could not be carried out due to the study's methodology. It should also not be forgotten that, by only analysing data for the years 2019, 2015 and 2010, the data found may be an underestimate or overestimate.

One of the strengths of this study is its originality, being one of very few studies to investigate the current situation and trend with regard to the publication of case reports in the scientific literature and, to the best of our knowledge, the first such study to date in endocrinology and nutrition journals. The fact that all of the case reports published over a considerable timespan and in a large number of endocrinology journals indexed in PubMed were analysed could also be considered an advantage.

To conclude, there is currently a falling trend in the publication of case reports in the scientific literature in general and in endocrinology in particular, both in absolute terms and in proportion to the total number of articles published. To the nest of our knowledge, this is the first study to cast light on this situation in this area of medical knowledge. This reduction in the publication of case reports may contribute to the loss of some clinical and scientific knowledge, as what is not published cannot be read by other authors nor serve as a basis for future research studying associations that might reach more scientifically robust conclusions. This type of scientific communication, where it concerns truly novel cases and is communicated in journals with reach and impact, should not therefore be forgotten.

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Conflicts of interest

The authors declare that they have no conflicts of interest.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j. endien.2022.02.007.

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