ESC European Society of Cardiology

European Heart Journal - Case Reports (2020) **4**, 1–5
doi:10.1093/ehjcr/ytaa251

ORIGINAL RESEARCH ARTICLE
Other

An assessment of adherence to CARE reporting standards by case reports published in *European Heart Journal – Case Reports* in 2018

Rosie Freer [1], Alexandra Rowett [2], and C. Fielder Camm [3]*

[1]Pembroke College, University of Oxford, Oxford, UK; [2]Keble College, University of Oxford, Oxford, UK; and [3]Cardiology Department, Royal Berkshire Hospital, Reading, UK

Received 31 May 2020; first decision 22 June 2020; accepted 8 July 2020; online publish-ahead-of-print 26 September 2020

Background Case reports are subject to significant variation in their content, and the absence of pertinent case details can limit their benefit to the medical community. To aid this, a reporting standard (CARE) has been developed. Case reports published in *European Heart Journal – Case reports* (*EHJ-CR*) are subject to specific checks by editors to confirm compliance with the CARE reporting standard. However, a degree to which case reports published by *EHJ-CR* comply with the CARE reporting standards has not been established.

Methods Case reports published in *EHJ-CR* during 2018 were reviewed for compliance with the CARE reporting standards. Two authors assessed each article for compliance with each of the 31 criteria.

Results In 2018, 130 case reports/series were published by *EHJ-CR*. The median number of CARE criteria achieved by each article was 21 (interquartile range 21–25) out of 31. CARE criteria with the highest adherence were timeline inclusion, a clear and well-referenced discussion, and declaration of competing interests, all present in 100% of articles. In contrast, some aspects were poorly adhered to including patient perspective, and details of funding sources. There was no difference in overall compliance with aspects of the CARE standard between diagnostic and interventional case reports. However, lower compliance was seen for the discussion of diagnostic challenges in interventional studies (19%), when compared to diagnostic studies (44%). The continent of authorship and month submitted did not affect CARE adherence.

Conclusions There was good compliance with the CARE reporting standards by case reports published in *EHJ-CR*. A number of specific areas for improvement have been identified which will be considered by the editorial board of *EHJ-CR*.

Keywords Case reports • CARE • Reporting standards • Audit

Introduction

Case reports are a common form of publication where a treating team describes a clinical case and places it into the wider context of scientific literature. Cases described are subject to significant publication bias and often represent extreme examples of pathology or very novel treatment.[1,2] Despite this, case reports remain important publication vehicles and have played significant historical roles for establishing new diseases, including Zika virus infection and AIDS,[3,4] and delineating important aspects of emerging conditions such as during the current COVID-19 pandemic.[5,6]

Due to the highly heterogeneous nature of case reports and often non-specialist first authors,[7] there is a risk that the cases will be poorly described. Case reports missing key clinical details will be of limited benefit to the medical community, and if misunderstood or misapplied can prove to be poor guides of medical practice.[8–11]

* Corresponding author. Email: Christian.camm@chch.ox.ac.uk
Handling Editor: Tom De Potter
Peer-reviewers: Richard Ang and Bogdan Enache
© The Author(s) 2020. Published by Oxford University Press on behalf of the European Society of Cardiology.
This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (http://creativecommons.org/licenses/by-nc/4.0/), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

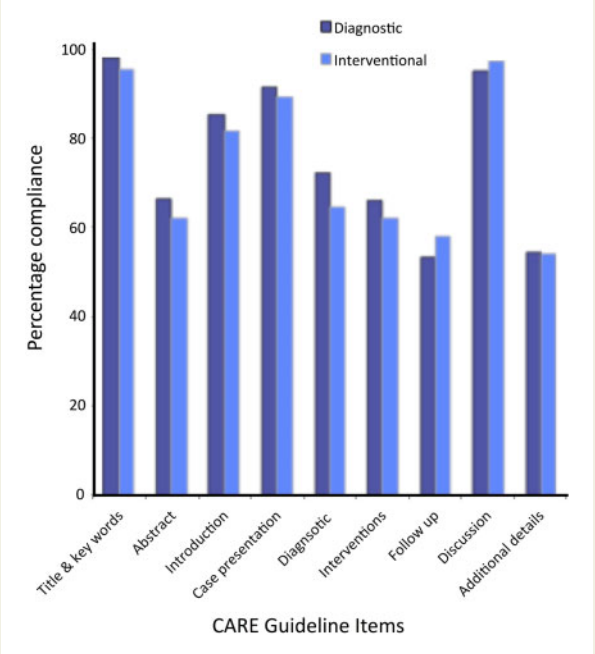

Figure 1 Compliance for diagnostic and interventional studies with the CARE reporting standard items grouped into key themes.

The need for improved reporting standards for case reports has been highlighted in several reviews.[12–14] To address this problem, the reporting standard CARE (CAse REport), has been established for Case Reports.[15]

European Heart Journal – Case reports (*EHJ-CR*) is an open-access journal specializing in the publication of cardiovascular case reports.[16] From its foundation, the editorial board has set out to maintain a high standard for the cases published in the journal. Thus, the case reports should conform to CARE reporting standards.[17] To achieve this, the journal has editors whose sole responsibility is to assess articles accepted for publication to confirm their adherence to these reporting standards.[18]

Manually checking accepted articles for compliance with reporting standards is a significant burden on the editorial team of *EHJ-CR*. To determine if the reporting standards are being met with this current process, we set out to assess articles published by *EHJ-CR* against the published CARE criteria.

Methods

Case selection

Cases were included if they were 'Case Reports' or 'Case Series' articles published by *EHJ-CR* and published in 2018. Article details were obtained from the journal website (https://academic.oup.com/ehjcr) with further meta-data obtained as necessary from the journal submission system. Articles were excluded if they were published in an issue before or after 2018 or if they were of another article type (other article types are not subjected to the same CARE reporting standard adherence check). Articles were divided based on their topic and whether focused on diagnostic or interventional aspects of the case.

Scoring against CARE reporting standard

Two authors (R.F. and A.R.) assessed each published report against the CARE reporting standard using the published checklist.[15] Prior to undertaking the scoring, both authors fully familiarized themselves with the reporting standard and undertook a trial assessment of five randomly selected articles from another time period. These trial articles were then discussed between the assessors and an editor who had previously acted as a CARE compliance reviewer for *EHJ-CR*.

After scoring each article, responses between authors were compared and differences resolved through consensus.

Statistical analysis

The Shapiro–Wilks test identified that the data were not normally distributed.[19] Medians and interquartile ranges were therefore used to describe the parametric data. The Mann–Whitney *U* test was used to compare summed CARE responses between case report diagnostic and interventional sub-categorizations. Multiple hypothesis corrections using the Benjamini–Hochberg method were used to adjust *P*-values appropriately when specific subsets of items were considered.[20] A *P*-value of <0.05 was considered significant. A Kruskal–Wallis *H* test was used to analyse variance between CARE reporting standard scoring grouped by reviewer, date of publication, location of authorship, and academic focus.[21] Agreement between reviewer assessment of case reports against CARE reporting standards was evaluated using Cohen's Kappa (κ) score.[22,23]

Results

A total of 159 articles were published by *EHJ-CR* during 2018 and 130 were case reports or case series, all of which were included in this analysis. The Cohen's κ score between reviewers was 0.63, with significant variation in Cohen's κ between items (Supplementary material online, *Table S1*). The largest proportion of publications were submitted from Europe (49%), followed by Asia (27%), and North America (15%). The majority of publications were case reports, with only 6% being case series. The cases described a range of cardiac diseases and imaging techniques. Full details of cases assessed are available in Supplementary material online, *Table S2*.

Out of 31 criteria, articles complied with a median of 23 (IQR 21–25). However, there was heterogeneity between criteria, with some aspects of the reporting standards being adhered to closely, whilst others were poorly addressed (*Table 1*). CARE reporting standards with the highest adherence were timeline inclusion (Item 7), a clear and well-referenced discussion (Items 11b–d) and declaration of competing interests (Item 13c), all present in 100% of articles. In contrast, adherence to other aspects was poor; only 6 articles (5%) included a patient perspective (Item 12), and only 17 articles (13%) confirmed details of funding sources or absence thereof (Item 13b).

To ascertain whether some instances of poor adherence were a consequence of items unsuitable for particular case reports, we calculated variation in compliance between diagnostic and interventional publications. Forty-three articles (33%) were felt to focus primarily on an interventional aspect, while 87 articles (67%) focused primarily on diagnostic aspects. There was no difference in overall compliance with aspects of the CARE standard between diagnostic and interventional case reports, with a median of 23 (IQR 22–25) for diagnostic and 22 (IQR 21–24) for interventional studies ($P = 0.1$). When CARE reporting standard items were grouped into key reporting themes,

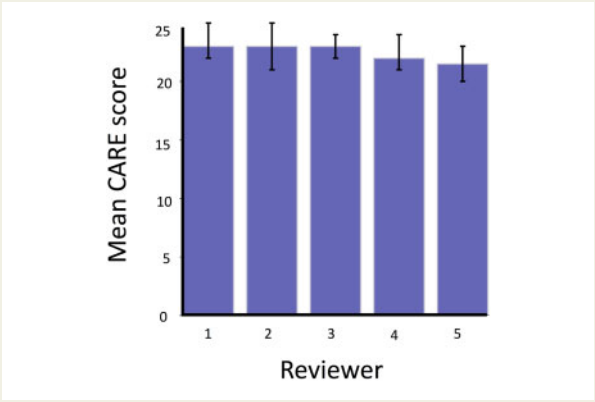

Figure 2 Median CARE compliance for articles broken down by *EHJ-CR* compliance editor. Maximum value 31. Error bars indicate the interquartile range.

there was good agreement between diagnostic and interventional articles (*Figure 1*). However, when individual care items were considered (Supplementary material online, *Figure S1*), lower compliance was seen for the discussion of diagnostic challenges in interventional studies (19%), when compared to diagnostic studies (44%) (Item 8b, $P = 0.02$). In addition, there was significantly better compliance regarding clinician-assessed outcomes (Item 10b, $P = 0.009$) for interventional (100%) than for diagnostic case reports (82%).

To confirm that reviews of compliance to CARE reporting standards were consistent, we stratified the results by the junior editor undertaking the review. There was no significant difference when articles were stratified by the member of the editorial board undertaking the compliance checks ($P = 0.2$, *Figure 2*).

We completed further consistency checks to ascertain whether unseen biases were impacting the quality of reports published in *EHJ-CR*. There was no significant variation in CARE compliance when each month throughout 2018 was compared ($P = 0.09$, Supplementary material online, *Figure S2A*). The variation between CARE compliance was also analysed for case report theme and the continent of authorship (Supplementary material online, *Figures S2B, C*). Here, we also established that there was no significant variation ($P = 0.2$ for report theme, $P = 0.3$ for the continent of authorship).

Discussion

Our study has shown good (74%) compliance with CARE reporting standards for case reports published in 2018 by *EHJ-CR*. Compliance to CARE standards was independent of the location of authors (by the continent), date of publication, and focus of the case report (case section). Reporting standards were consistent across *EHJ-CR* reviewers, indicating systematic assessment against CARE standards of all papers submitted to the journal.

A substantial number of reporting standards have been developed for health research, with the aim of improving the reliability and value of published work. The EQUATOR Network contains details of 424 such standards.[24] There is evidence that the use of standards promotes high-quality reporting. A Cochrane review found more complete reporting in CONSORT standard-endorsing journals for 25 of 27 assessed outcomes.[25] However, data from Turner *et al*. also support our finding that the endorsement of reporting standards does not ensure 100% compliance with all desired outcomes.[25]

Compliance may vary between reporting standards for several reasons. Some items, such as the inclusion of a timeline (Item 7, 100% compliance), can be objectively assessed and were required prior to publication. Other items are more subjective, such as discussion of case management limitations (Item 11a, 82% compliance).

The subjective nature of many CARE items may underlie the relatively low agreement between reviewers (Cohen's κ score = 0.63) when compared to previous reviews of publications using other reporting standards (Cohen's κ score = 0.74–0.93).[26–29] Assessment of reviewer agreement for individual items (Supplementary material online, *Table S1*), may identify reporting standards which could benefit from improved objectivity. For instance, Item 3c asked reviewers to judge the quality of the Abstract conclusion. Heterogeneity between reviewer's scoring (Cohen's κ = -0.027), revealed this item to be open to subjective opinion.

Identifying items with the poorest agreement between reviewers and discussing varying interpretations amongst assessors may inform changes to the CARE reporting standard, enabling more consistent assessment. As CARE guidelines are used by a number of other journals in addition to *EHJ-CR*, it may be useful to also assess the CARE compliance within and between other journals to inform reporting standard changes. Improved clarity within items would ensure that both authors and reviewers are able to accurately judge the manuscript against the reporting standard. However, subjective interpretation of the reporting standard cannot completely account for instances of poor compliance, with potential for improvement found in some items with good inter-reviewer concordance (Items 12 and 13b; *Table 1* and Supplementary material online, *Table S1*).

In instances of high reviewer agreement, compliance can be improved by increased adherence to reporting standard criteria amongst study authors and reviewers. However, guideline authors must also ensure that the reporting standards created set appropriate requirements for the article type. In our study, we found significantly lower CARE compliance for the reporting of diagnostic challenges in case reports with an interventional focus. We also identified significantly reduced reporting of clinician-assessed outcomes for case reports with a diagnostic focus. It may be that the creation of two distinct CARE reporting standards—for diagnostic and interventional studies—would allow authors to comply with guidelines whilst reporting only the most pertinent information. There are some reporting standards that may be inappropriate for both diagnostic and interventional studies. A patient's perspective can add depth to a case report, but it is not always possible to gather a patient's insight into their care in a meaningful way. Because of this, *EHJ-CR* does not insist on the inclusion of patient perspective in publications.

In other instances, it is important for authors to comment on items, whether the case can provide the information requested or not. One such example is Item 10d, which asks authors to report adverse or unanticipated events, or else explicitly mention their absence. Only 38% of publications did so. In response, the *EHJ-CR* will

Table 1 Adherence to CARE reporting standard items by case reports published in 2018

Topics	Item	Checklist item description	*n* (%)
Title	1	Must contain the words 'case report', 'case study', or 'case series'	129 (99)
Key words	2	3–7 key words that identify areas covered in the case report (include 'case report/study/series' as one of the key words)	123 (95)
Abstract	3a	Introduction—what is unique or educational	90 (69)
	3b	Case summary, including; symptoms, clinical findings, diagnosis, interventions, and outcomes	38 (29)
	3c	Conclusion	125 (96)
Introduction	4	A brief background summary of the case, referencing medical literature	109 (84)
Patient information	5a	De-identified demographic and patient-specific information	127 (98)
	5b	Presenting complaint including the timing of symptoms if appropriate	89 (68)
	5c	Relevant past medical and social history	122 (94)
Clinical findings	6	Relevant physical examination findings	121 (93)
Timeline	7	Inclusion of data in a table which establishes the sequence of events which comprise the patient's history and presentation	130 (100)
Diagnostic assessment	8ai	Diagnostic methods (including lab testing, imaging, surveys)	129 (99)
	8aii	Normal values for any reported diagnostic studies reported in brackets where appropriate	126 (97)
	8b	Diagnostic challenges (including financial, language, cultural, and technical)	46 (35)
	8c	Diagnostic reasoning, including other diagnoses considered	71 (55)
	8d	Prognostic characteristics	81 (62)
Therapeutic intervention	9a	Intervention type clearly stated (can include pharmacological, surgical, and preventative)	129 (99)
	9b	Details of how the intervention was administered, including dosage, strength, and duration	56 (43)
	9c	Any changes in intervention with rationale	67 (52)
Follow-up and outcomes	10a	Patient-reported outcomes	101 (78)
	10b	Clinician-assessed outcomes	114 (88)
	10c	Follow-up intervention and tolerability to the main intervention, with compliance specifically discussed	20 (15)
	10d	Adverse and unanticipated events as a result of any intervention	50 (38)
Discussion	11a	Strengths and limitations of the management of this case	107 (82)
	11b	Discussion of relevant medical literature	130 (100)
	11c	The rationale for any conclusions made	130 (100)
	11d	The primary 'take away' lessons from the case report, stated explicitly	130 (100)
Additional details	12	Detail the patient's perspective or experience	6 (5)
	13a	Confirmation of informed patient consent. Where not possible, an explanation of why and alternative arrangements should be detailed.	129 (99)
	13b	Details of funding sources	17 (13)
	13c	Presence or absence of conflict of interest confirmed	130 (100)

re-iterate the need to comment on items, even if negative, to both authors and compliance team reviewers.

For some items, a change in publication format by *EHJ-CR* would improve compliance. This is applicable in particular for funding sources (Item 13b). Information on potential financial conflicts is already collected from authors. At present, the journal only reports this information when work is funded. As most case reports are not funded, there is currently a low level of compliance with this reporting standard. However, *EHJ-CR* will now include a funding statement in all its articles, ensuring 100% compliance in the future for this item.

In addition to the creation of more tailored reporting standards and publication protocols, interventions to improve reporting quality should include further compliance training and surveillance by *EHJ-CR*. Editors will be informed of areas of poor compliance during training, and a regular audit process is being developed within *EHJ-CR* to provide feedback to editors in a timely manner. It would be useful to complete a follow-up study subsequent to these interventions to assess for improved reporting standards and consistency between *EHJ-CR* editors.

Conclusion

Case reports published in *EHJ-CR* in 2018 show good (74%) compliance to CARE reporting standards. Differences in compliance between diagnostic and interventional case reports highlight a need for more tailored reporting standards. An opportunity to improve reporting of potential financial conflicts has been identified, and

EHJ-CR will amend protocols to ensure these are stated in all instances. *EHJ-CR* will also implement further compliance training and surveillance, using CARE as a tool to improve the reporting quality of publications.

Lead author biography

Fielder Camm is the Assistant Editor of *European Heart Journal - Case Reports* and a Cardiology Trainee. His areas of interest including arrhythmias and big data. In addition, he is passionate about reporting standards in academic publications

Supplementary material

Supplementary material is available at *European Heart Journal - Case Reports* online.

Funding

No specific funding was used for this research project.

Conflict of interest: Dr Camm is Assistant Editor for *European Heart Journal - Case Reports*. This manuscript was reviewed in a double blind manner and Dr Camm was not involved in the editorial or review process.

References

1. Easterbrook PJ, Gopalan R, Berlin JA, Matthews DR. Publication bias in clinical research. *Lancet* 1991;**337**:867–872.
2. Albrecht J, Meves A, Bigby M. Case reports and case series from Lancet had significant impact on medical literature. *J Clin Epidemiol* 2005;**58**:1227–1232.
3. Macnamara F. Zika virus: a report on three cases of human infection during an epidemic of jaundice in Nigeria. *Trans R Soc Trop Med Hyg* 1954;**48**: 139–145.
4. Simpson D. Zika virus infection in man. *Trans R Soc Trop Med Hyg* 1964;**58**: 339–338.
5. Lescure F-X, Bouadma L, Nguyen D, Parisey M, Wicky P-H, Behillil S, et al. Clinical and virological data of the first cases of COVID-19 in Europe: a case series. *Lancet Infect Dis* 2020;**20**:697–706.
6. Xu Z, Shi L, Wang Y, Zhang J, Huang L, Zhang C et al. Pathological findings of COVID-19 associated with acute respiratory distress syndrome. *Lancet Respir Med* 2020;**8**:420–422.
7. Lundh A, Christensen M, Jørgensen AW. International or national publication of case reports. *Dan Med Bul* 2011;**58**:A4242.
8. Wangensteen OH, Salmon PA, Griffen WO, Paterson JR, Fattah F. Studies of local gastric cooling as related to peptic ulcer. *Ann Surg* 1959;**150**:346–360.
9. Belhassen B, Rotmensch HH, Laniado S. Response of recurrent sustained ventricular tachycardia to verapamil. *Heart* 1981;**46**:679–682.
10. Snyder BD, Blonde L, McWhirter WR. Reversal of amitriptyline intoxication by physostigmine. *JAMA* 1974;**230**:1433–1434.
11. Crile G, *Blood Pressure in Surgery, Experimental and Clinical Research.* Philadelphia: Lippincott Company, 1903.
12. Kaszkin-Bettag M, Hildebrandt W. Case reports on cancer therapies: the urgent need to improve the reporting quality. *Glob Adv Health Med* 2012;**1**:8–10.
13. Kljakovic M. Single cases in general practice and general medical journals. *Aust Fam Phys* 2002;**31**:669–673.
14. Richason TP, Paulson SM, Lowenstein SR, Heard KJ. Case reports describing treatments in the emergency medicine literature: missing and misleading information. *BMC Emerg Med* 2009;**9**:10.
15. Gagnier JJ, Kienle G, Altman DG, Moher D, Sox H, Riley D; the CARE Group. The CARE guidelines: consensus-based clinical case reporting guideline development. *J Med Case Rep* 2013;**7**:223.
16. Camm CF, Camm AJ. European Heart Journal-Case Reports (EHJ-CR): a new format for an old concept. *Eur Heart J Case Rep* 2017;**1**: 1–2.
17. https://academic.oup.com/ehjcr/pages/General_Instructions (20 April 2020).
18. https://academic.oup.com/ehjcr/pages/editor_and_reviewer_programmes (20 April 2020).
19. Shapiro SS, Wilk MB. An analysis of variance test for normality (complete samples). *Biometrika* 1965;**52**:591–611.
20. Benjamini Y, Hochberg Y. Controlling the false discovery rate: a practical and powerful approach to multiple testing. *J R Stat Soc B* 1995;**57**:289–300.
21. Kruskal WH, Wallis WA. Use of ranks in one-criterion variance analysis. *J Am Stat Assoc* 1952;**47**:583–621.
22. Cohen J. A coefficient of agreement for nominal scales. *Educ Psychol Measure* 1960;**20**:37–46.
23. Cohen J. Weighted kappa: nominal scale agreement provision for scaled disagreement or partial credit. *Psychol Bull* 1968;**70**:213–220.
24. Equator network [online]. http://www.equator-network.org/.
25. Turner L, Shamseer L, Altman DG, Weeks L, Peters J, Kober T et al. Consolidated standards of reporting trials (CONSORT) and the completeness of reporting of randomised controlled trials (RCTs) published in medical journals. *Cochrane Database Syst Rev* 2012;**11**:MR000030.
26. Camm CF, Chen Y, Sunderland N, Nagendran M, Maruthappu M, Camm AJ. An assessment of the reporting quality of randomised controlled trials relating to anti-arrhythmic agents (2002–2011). *Int J Cardiol* 2013;**168**:1393–1396.
27. Agha RA, Camm CF, Edison E, Orgill DP. The methodological quality of randomized controlled trials in plastic surgery needs improvement: a systematic review. *J Plastic Reconstruct Aesthet Surg* 2013;**66**:447–452.
28. Agha RA, Camm CF, Doganay E, Edison E, Siddiqui MRS, Orgill DP. Randomised controlled trials in plastic surgery: a systematic review of reporting quality. *Eur J Plast Surg* 2014;**37**:55–62.
29. Lee S-Y, et al. Compliance of randomized controlled trials in trauma surgery with the CONSORT statement. *J Trauma Acute Care Surg* 2013;**75**:562–572.