Introduction: Case series have been a long held tradition within the surgical literature and are still frequently published. Reporting guidelines can improve transparency and reporting quality. No guideline exists for reporting case series, and our recent systematic review highlights the fact that key data are being missed from such reports. Our objective was to develop reporting guidelines for surgical case series.

Methods: A Delphi consensus exercise was conducted to determine items to include in the reporting guideline. Items included those identified from a previous systematic review on case series and those included in the SCARE Guidelines for case reports. The Delphi questionnaire was administered via Google Forms and conducted using standard Delphi methodology. Surgeons and others with expertise in the reporting of case series were invited to participate. In round one, participants voted to define case series and also what elements should be included in them. In round two, participants voted on what items to include in the PROCESS guideline using a nine-point Likert scale to assess agreement as proposed by the Grading of Recommendations, Assessment, Development and Evaluations (GRADE) working group.

Results: In round one, there was a 49% (29/59) response rate. Following adjustment of the guideline with incorporation of recommended changes, round two commenced and there was an 81% (48/59) response rate. All but one of the items were approved by the participants and Likert scores 7-9 were awarded by >70% of respondents. The final guideline consists of an eight item checklist.

Conclusion: We present the PROCESS Guideline, consisting of an eight item checklist that will improve the reporting quality of surgical case series. We encourage authors, reviewers, editors, journals, publishers and the wider surgical and scholarly community to adopt these.

© 2016 IJS Publishing Group Ltd. Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).
between case series and single group cohort studies [2].

As with case reports, their value has been debated [3,4]. In the age of evidence-based medicine (EBM), with the randomised controlled trial as the standard to show the efficacy of a particular treatment, what is their role? Level four evidence was still the most common study type in a bibliometric analysis of research published in 2013 in the specialties of plastic surgery, orthopaedic surgery, otolaryngology and neurosurgery, with significant outputs in maxillo-facial surgery (33%) and vascular surgery (15%) [5]. The use of a case series in the recognition of a new disease was exemplified in 1999 by the epidemic of West Nile encephalitis in New York [6]. Historically, case series were important in identifying the impact of maternal drinking and pregnancy outcome and the role of vitamin C in preventing scurvy [7,8]. A single case series can lead to very significant change, from the widespread use of negative pressure dressings following a case series of 10 patients [9] to a 49 patient case series that led to a new classification system for haemangiomas and vascular malformations in 1982, still in use today [10].

Albrecht et al. studied reports of case series and found that a high proportion led to follow-up trials and that they were useful in establishing an early evidence base for new treatments of rare diseases in which trials would not be feasible [11]. For some specialties, establishing control groups may be difficult, such as in accident and emergency medicine or paediatric medicine or surgery. In the social sciences, many social psychology studies have been case series, for example Yale psychologist Stanley Milgram’s seminal work on obedience to authority figures [12].

In a 2005 report, Dalziel et al. found that case series were used in 30% of Health Technology Assessments (HTA) used in the provision and suitability of care [13]. Poor reporting in the case series included in their study, however, severely constrained their analysis and investigation of the hypothesis that findings in case series may be affected by methodological characteristics [10]. Readers need complete, transparent information in all reports of research. Poor reporting of case series undermines critical appraisal, assessment of external validity and whether, for instance, surgeons should change their practice.

No standardised reporting criteria have been developed within a robust methodological framework for case series. The aim of the present study was to close this gap and produce a reporting guideline for case series that is methodologically robust, easy to use, and accepted internationally across a broad range of specialties and disciplines. Following guidance on guideline development, the early steps in this process require an analysis of previous literature to identify previous guidance (if any) and to analyse relevant evidence on the quality of reporting of published research articles within the domain of interest [14]. Our group recently completed a systematic review on the reporting quality of case series in surgery over the period 1990–2014 [15]. From 92 articles that met the inclusion criteria, methodological and reporting issues identified were: failure to use standardised definitions (57%), missing or selective data (66%), lack of transparency or incomplete reporting (70%), whether alternative study designs were considered (11%) and other issues (52%) such as failure to clearly define the patient population under investigation, selection bias, insufficient follow-up time, need for validated outcomes.

We recently developed the SCARE Guidelines for Case Reports using a DELPHI consensus exercise, which have now been adopted by several journals [16]. Following this experience, the objective of this research was to conduct a Delphi consensus exercise amongst experienced surgical case series reviewers and editors to develop the Preferred Reporting Of Case Series in Surgery (PROCESS) Guideline.

2. Developing the PROCESS guideline

We issued a survey using Google Forms (https://www.google.co.uk/forms/about) asking participants in round one to help define surgical case series and what items should be included in them. In a subsequent round, participants were asked to rate their level of agreement with the guideline items from round one as well as items from the SCARE guidelines and any additional items that were suggested using a nine-point Likert scale as proposed by the GRADE group [17]. In this scale 1 to 3 signifies an outcome of limited importance, 4 to 6 important but not critical and 7 to 9 critical. If 70% or more of respondents scored an item 7 to 9 and fewer than 15% scored it 1 to 3, that item was incorporated in the reporting guideline. Similarly, consensus that an outcome should not be included was 70% or more scoring it 1 to 3 and 15% or less scoring it 7 to 9. The entire process was conducted electronically and there was no pre-determined number of Delphi rounds.

3. Participant selection

Surgeons and others with significant experience in reviewing or editing case reports were selected. They were drawn from the reviewer pool of IJS Case Reports (the top 150 were invited) as well as those who have written on the topic of case series and case reports in the past. In total 59 participants agreed to the invitation to participate in this study, representing 21 countries and all ten surgical specialties as well as allied specialties including; dermatology, pathology, oncology, clinical pharmacology, acute care surgery, with many participants also occupying positions on journal editorial boards [18].

4. Results

In round 1, there was a 49% (29/59) response rate. The participant responses are integrated into Table 1. Following adjustment of the guideline with incorporation of recommended changes, round 2 commenced (see Tables 2 and 3). There was an 81% (48/59) response rate. All guideline items were approved by the participants with Likert scores 7–9 awarded by >70% of respondents, apart from “4a - registration and ethics - state the research registry number in accordance with the declaration of Helsinki”, which had scores of 7–9 from 65% of participants. However, as this item is part of the declaration of Helsinki, it cannot be removed or augmented.

5. PROCESS guideline

Table 3 constitutes the PROCESS guideline, and this is provided again in an Appendix, together with a column in which the author can state the page number on which the criterion was achieved. We recommend that all authors submitting case series should submit a completed PROCESS checklist with their manuscript and also state explicitly in their report that they have complied with the PROCESS guideline, which they should cite in their paper. The guideline represents the minimum of what should be reported and we encourage authors to provide additional details that are relevant. So, when should a case series be performed and when should these guidelines be used? For surgical case series specifically, the following can also be advocated: rare diseases or rare circumstances (such as emergencies), logistical difficulties or where ethical issues may arise with prospective randomised studies e.g. paediatric populations, new diseases – their description, natural history and management, studying the mechanism of disease and studying the impact of established procedures. In addition, late or delayed effects following surgical interventions, such as biliary
malignancy after biliodigestive anastomosis, could be collated into a case series. Where a new technique or device has been conceived and requires development and assessment, the IDEAL (Idea, Development, Exploration, Assessment and Long-term follow-up) framework is recommended [19].

6. Endorsement

The PROCESS guideline has been endorsed by the IJS, IJS Case Reports, IJS Open, Annals of Medicine and Surgery, IJS Oncology, and IJS Short Reports.

7. Conclusion

After completion of two DELPHI rounds consensus was reached among a multidisciplinary and expert group in the area of surgery and case series. If used appropriately, the PROCESS guidelines will aid in raising the reporting quality of surgical case series. We encourage authors, reviewers, editors, journals, publishers and the wider surgical and scholarly community to adopt these. We look forward to feedback from the community as well as studies of its implementation to help inform a future revision of these guidelines.

8. PROCESS group participants

The following people contributed to the PROCESS Guideline: Raafat Afffi, Cairo University, Raha Alahmadi, King Faisal Specialist Hospital and Research Centre, Joerg Albrecht, John H. Strorger Jr. Hospital of Cook County, Abdulrahman Alsawadi, Colchester Hospital University NHS Foundation Trust, Jeffrey K. Aronson, Radcliffe Infirmary, Oxford, M. Hammad Ather, Aga Khan University, Mohammad Bashashati, Texas Tech University Health Sciences Centre, Somprakas Basu, Banaras Hindu University, Patrick Bradley, Nottingham University Hospitals, Mushtaq Chalkoo, Govt. medical college, Srinagar Kashmir, Ben Challacombe, Guy’s and St Thomas’ NHS Foundation Trust, Trent Cross, James Cook University, Laura Derbyshire, North West Deanery, Naheed Farooq, Central Manchester University Hospital Foundation Trust, Jerome Hoffman, University of California Los Angeles, Huseyin Kadioglu, Bezmialem Vakif University, Veeru Kasivisvanathan, University College London, Boris Kirkstein, Soroka University Medical Centre, Roberto Kloppenbach, Simplemente Evita Hospital, Daniel Laskin, Virginia Commonwealth University, Diana Miguel, University Hospital Jena, James Milburn, Queens Medical Centre, Oliver Muensterer, University Medicine Mainz, James Ngu, Changi General Hospital, Iain Nixon, East Kent University Hospitals, Ashraf Noureldin, Cumberland Royal Infirmary, Benjamin Perakath, Dr. Gray’s Hospital.
### Table 3
**DELPHI round 2 responses to guideline items.**

<table>
<thead>
<tr>
<th>Section</th>
<th>Item</th>
<th>Checklist Description</th>
<th>Responses (n = 48)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Title</strong></td>
<td>1</td>
<td>The words “case series” and the area of focus should appear in the title (e.g., disease, exposure/intervention or outcome).</td>
<td>80% (37/46)</td>
</tr>
<tr>
<td><strong>Abstract</strong></td>
<td>2a</td>
<td><strong>Introduction</strong></td>
<td>89% (41/46)</td>
</tr>
<tr>
<td></td>
<td>2b</td>
<td><strong>Methods</strong></td>
<td>89% (41/46)</td>
</tr>
<tr>
<td></td>
<td>2c</td>
<td><strong>Results</strong></td>
<td>96% (44/46)</td>
</tr>
<tr>
<td></td>
<td>2d</td>
<td><strong>Conclusion</strong></td>
<td>65% (30/46)</td>
</tr>
<tr>
<td><strong>Introduction</strong></td>
<td>3</td>
<td>Explain the scientific background and rationale for the case series. What is the unifying theme - common disease, exposure, intervention and outcome, etc. Why is this study needed?</td>
<td>91% (42/46)</td>
</tr>
<tr>
<td><strong>Methods</strong></td>
<td>4a</td>
<td><strong>Registration and ethics</strong></td>
<td>87% (40/46)</td>
</tr>
<tr>
<td></td>
<td>4b</td>
<td><strong>Study design</strong></td>
<td>93% (43/46)</td>
</tr>
<tr>
<td></td>
<td>4c</td>
<td><strong>Setting</strong></td>
<td>93% (43/46)</td>
</tr>
<tr>
<td></td>
<td>4d</td>
<td><strong>Participants</strong></td>
<td>e.g. state why.</td>
</tr>
<tr>
<td></td>
<td>4e</td>
<td><strong>Pre-intervention considerations</strong></td>
<td>80% (37/46)</td>
</tr>
<tr>
<td></td>
<td>4f</td>
<td><strong>Types of intervention(s) deployed</strong></td>
<td>87% (40/46)</td>
</tr>
<tr>
<td></td>
<td>4g</td>
<td><strong>Peri-intervention considerations</strong></td>
<td>89% (41/46)</td>
</tr>
<tr>
<td></td>
<td>4h</td>
<td><strong>Who performed the procedure(s)</strong></td>
<td>83% (38/46)</td>
</tr>
<tr>
<td><strong>Results</strong></td>
<td>5a</td>
<td><strong>Participants</strong></td>
<td>96% (44/46)</td>
</tr>
<tr>
<td></td>
<td>5b</td>
<td><strong>Changes</strong></td>
<td>89% (41/46)</td>
</tr>
<tr>
<td></td>
<td>5c</td>
<td><strong>Outcomes and follow-up</strong></td>
<td>93% (43/46)</td>
</tr>
<tr>
<td></td>
<td>5d</td>
<td><strong>Intervention adherence/compliance and tolerability</strong></td>
<td>91% (42/46)</td>
</tr>
<tr>
<td></td>
<td>5e</td>
<td><strong>Complications and adverse or unanticipated events</strong></td>
<td>89% (41/46)</td>
</tr>
<tr>
<td><strong>Discussion</strong></td>
<td>6a</td>
<td><strong>Summarise key results</strong></td>
<td>93% (43/46)</td>
</tr>
</tbody>
</table>
Nicholas Raison, King’s College London, Kandiah Raveendran, Fatimah Hospital, Timothy Sullivan, Minneapolis Heart Institute, Achilles Thoma, McMaster University, Mangesh A. Thorat, Wolfson Institute of Preventive Medicine, Queen Mary University of London, Andy Petroianu, Federal University of Minas Gerais, Ashwini Rao, Manipal College of Dental Sciences, Mangalore, Manipal University, Michele Valmasoni, Università di Padova, Samuele Massarut, Centro di Riferimento Oncologico Aviano Italy, Anil D’cruz, Tata Memorial Hospital, Baskaran Vasudevan, MIOT Hospitals, Salvatore Giordano, Turku University Hospital, Donagh Healy, University Hospital Waterford, David Machado-Aranda, University of Michigan, Frederick H. Millham, Newton-Wellesley Hospital, Bryan Carrol, Eastern Virginia Medical School, Indraneilm Mukherjee, Florida Hospital Tampa, Peter McCulloch, University of Oxford, Yasuhiiko Sugawara, Japanese Red Cross Hospital and David Rosin, University of West Indies.

Ethical approval
Not applicable.

Sources of funding
Paton Masser Memorial Fund Prize awarded by British Association of Plastic, Reconstructive and Aesthetic Surgeons and Balliol Interdisciplinary Institute Grant. None of the authors has a financial interest in any of the products, devices or drugs mentioned in this manuscript.

Author contribution
Authors:
Riaz A. Agha: Concept and design of study, data collection, data interpretation and analysis, drafting, revision, approval of final manuscript.
Alexander J Fowler: Study design, data collection, revision, approval of final manuscript.
Shivanchan Rajmohan: Data Collection, revision, approval of final manuscript.
Ishani Barai: Data collection, revision, approval of final manuscript.
Dennis P. Orgill: Design of study, revision, approval of final manuscript.

Conflicts of interest
None declared. The authors have no financial, consultative, institutional, and other relationships that might lead to bias or conflict of interest.

Guarantor
Riaz. A. Agha.

Appendix A. Supplementary data
Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.ijsu.2016.10.025.

References