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PRICE 2020 Guidelines for reporting case reports in Endodontics: A consensus-based development

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Abstract

Case reports can provide early information about new, unusual or rare disease(s), newer treatment strategies, improved therapeutic benefits and adverse effects of interventions or medications. This paper describes the process that led to the development of the Preferred Reporting Items for Case reports in Endodontics (PRICE) 2020 guidelines through a consensus-based methodology. A steering committee was formed with eight members (PD, VN, BC, PM, PS, EP, JJ, SP), including the project leaders (PD, VN). The steering committee developed an initial checklist by combining and modifying the items from the Case Report (CARE) guidelines and Clinical and Laboratory Images in Publications (CLIP) principles. A PRICE Delphi Group (PDG) and PRICE Face-to-Face Meeting Group (PFMG) were then formed. The members of the PDG were invited to participate in an online Delphi process to achieve consensus on the wording and utility of the checklist items and the accompanying flowchart that was created to complement the PRICE 2020 guidelines. The revised PRICE checklist and flowchart developed by the online Delphi process was discussed by the PFMG at a meeting held during the 19th European Society of Endodontology (ESE) Biennial Congress in Vienna, Austria, in September 2019. Following the meeting, the steering committee created a final version of the guidelines, which were piloted by several authors during the writing of a case report. In order to help improve the clarity, completeness and quality of case reports in Endodontics, we encourage authors to use the PRICE 2020 guidelines.

Keywords

Case reports, consensus, Endodontics, guideline

Introduction

Case reports can be used to report new or rare disease(s), unusual condition(s), more modern management approaches or novel treatment techniques for the benefit of clinicians, including those with an interest in the specialty, as well as the broader scientific community. They can also be used to generate new and innovative hypotheses that can direct further research and clinical practice (Danish *et al.* 2017) and/or inform patient management guidelines to enhance treatment outcomes (Cohen 2006). In addition, case reports can provide early information about the therapeutic benefits, adverse/side-effects, and financial implications of interventions (Nayak 2010, Riley *et al.* 2017). In the context of medical decision making, case reports may have low specificity, but high sensitivity for detecting novelty (Vandenbroucke 2001).

There is general consensus that reporting guidelines are important for improving the quality of medical research (Wang *et al.* 2015). The CAse REport (CARE) guidelines (Gagnier *et al.* 2013) were developed through a consensus process, aimed at improving the reporting of information in case reports within the field of Medicine. The CARE guidelines direct authors to submit accurate, complete and transparent manuscripts when describing case reports and have been endorsed by many journals. The CARE guidelines consist of 13 items (domains) including the title, keywords, abstract, introduction, patient information, clinical findings, timeline, diagnostic assessment, therapeutic interventions, follow-up and outcomes, discussion, patient perspective, and informed consent.

The quality of case reports submitted in the field of Endodontics can be variable and is often sub-optimal; indeed, many are incomplete and inaccurate, and are consequently rejected for publication (Dummer PMH, unpublished data). It is logical therefore that guidelines for writing case reports in Endodontics will help authors to prepare accurate and complete reports. In addition, clinical photographs, radiographs and/or other images, e.g. histopathological sections, are often central to the description of a case, its management or the treatment outcome. Hence, case reports published in Endodontics must be accompanied by high-quality images. The Clinical and Laboratory Images in Publications (CLIP) principles (Lang et al. 2012)

were developed as guidance for the reporting of images; to provide readers with the information needed to assess the accuracy, validity, completeness and credibility of the interpretation and implications of images published in journals. Thus, adherence to the CLIP principles will improve the reporting quality of images and the accuracy of the information provided.

The Preferred Reporting Items for Case reports in Endodontics (PRICE 2020) guidelines were specifically developed by integrating and adapting the CARE guidelines (Gagnier *et al.* 2013) and CLIP principles (Lang *et al.* 2012). The guidelines are designed to help authors improve the completeness, accuracy and transparency of their case reports, thereby reducing the number of poorly composed manuscripts submitted to journals. Hence, the aim of this project was to develop the PRICE 2020 guidelines for case reports in the specialty of Endodontology through a well-documented consensus-based process.

Methods

The study design was approved by the Institutional Review Board on Research and Ethics of the International Medical University (IMU), Malaysia (No: IMU 450/2019). The development of the PRICE 2020 guidelines followed the Guidance for Developers of Health Research Reporting Guidelines (Moher *et al.* 2010). The protocol used to develop of the guidelines has been published (Nagendrababu *et al.* 2019).

Forming the steering committee and developing the initial PRICE 2020 checklist and flowchart

The project leaders (VN, PD) carried out a comprehensive literature search and concluded that guidelines for the reporting of cases in Endodontics were necessary. A steering committee consisting of eight members (PD, VN, BC, PM, PS, EP, JJ, SP), including the project leaders, was formed. A draft checklist, specifically relevant to the field of Endodontics, was created by combining and modifying the items from the CARE guidelines (Gagnier *et al.* 2013) and CLIP principles (Lang *et al.* 2012).

Online Delphi survey

The steering committee formed a PRICE Delphi Group (PDG) of 30 individuals that included 22 academics, four Endodontists, two general dentists and two patient representatives. Apart from the patient representatives, at least one of the following criteria had to be fulfilled to become a member of the PDG:

- published at least one case report related to Endodontics;
- published a manual, handbook, or method guidelines related to case reports in Endodontics;
- published any reporting guidelines for *in vitro/in vivo* research;
- a minimum of 15 years of clinical experience in dentistry.

The steering committee identified and invited the 30 individuals to participate in the online Delphi process. The invitation letter explained the need for reporting guidelines for case reports in Endodontics, described the process to be followed by the Delphi panel and the tasks expected of members. After receiving confirmation of their participation, a Delphi document, setting out the background to the process, the need for PRICE guidelines, the draft PRICE checklist, and a description of the online Delphi survey that explained the process for including/excluding items were shared with each member of the PDG. An iterative approach was employed using online surveys to gain consensus. The PDG members scored and gave their views on the suitability and clarity of each item of the draft PRICE checklist. For each item, the members were asked to give their opinion on whether the individual item was clear ('yes' or 'no') and should be included using a 9-point rating Likert scale (1 = 'definitely not include' to 9 = 'definitely include'). Members were also given the opportunity to express their opinion, in the form of free text, on the wording and their understanding of each item (Maher et al. 2015). For inclusion in the final list, items had to achieve a score between 7 and 9 by \geq 70%, and between 1 and 3 by \leq 30% of the members.

Similarly, items were excluded from the draft PRICE checklist if $\geq 70\%$ of members scored an item between 1 and 3, and $\leq 30\%$ of members scored it between 7 and 9. PDG members were also asked to provide their views on whether the flowchart was clear ('yes' or 'no'). When an item required modification, members

were asked to re-rate the revised version through subsequent surveys. The Delphi process continued until a consensus was reached and a final set of items was agreed upon by the PDG members (Agha *et al.* 2017). At the end of each round, the results of the Delphi process and the collective scores given by the individual members were shared. The responses were anonymised to ensure the comments from individuals were provided without pressure or influence. The revised PRICE checklist and flowchart created by the online Delphi process was then discussed during a PRICE Face-to-Face meeting.

Face-to-Face meeting

The steering committee organised a face-to-face meeting at the 19th European Society of Endodontology (ESE) Biennial Congress held in Vienna, Austria on 13th September 2019. The steering committee identified and contacted 20 individuals via email to make-up the PRICE Face-to-Face Meeting Group (PFMG). The eligibility criteria for the PFMG were the same as for the PDG. In addition, two postgraduate students in Endodontology were invited to share their views. After receiving confirmation of their agreement to participate, the PFMG was informed of the venue, date and time of the face-to-face meeting. The project leaders shared the draft PRICE checklist, flowchart, results of the Delphi process, list of members, and meeting agenda with the PFMG before the meeting.

At the meeting, the project leaders (PD, VN) presented the results of the online Delphi process, the rationale for including the items and the flow chart that accompanied the PRICE checklist. The PFGM discussed and shared their views on the PRICE checklist and flow chart.

Post-meeting activities

Based on the comments from the face-to-face meeting, the PRICE 2020 checklist and flowchart were finalised by the steering committee.

Results

Online Delphi process

In total, 30 individuals participated in the Delphi process. A 100% response rate was achieved in both Rounds 1 and 2. Round 1 consisted of a PRICE checklist with 12 sections and 47 individual items and a PRICE Flowchart. Out of the 47 items, 41 received a score between 7 and 9 by ≥70% of members. Therefore, they were retained in the PRICE checklist. The remaining 6 items and the flowchart were revised based on the comments received from the PDG and required further discussion. Thus, Round 2 consisted of a PRICE checklist with these 6 items and the revised PRICE flowchart. Among the 6 items, four items were awarded a score between 7 and 9 by ≥70% of members and were retained in the PRICE checklist. Two items remained controversial, and hence, consensus was not achieved. Several constructive comments were received from the PDG regarding the flowchart, which was revised accordingly.

Face-to-Face meeting

The two controversial items were included for discussion in the face-to-face meeting to determine their inclusion or exclusion from the PRICE checklist; the flowchart was also discussed. The PFMG decided to include both items (Patient perspective and Quality of images used in the case report – The circumstances (conditions) under which the image(s) were viewed and evaluated by the authors must be provided in the text) in the PRICE checklist with modifications. In addition, three items were removed from the PRICE checklist as they were deemed to be unnecessary and/or duplications of other items; three new items (ethnicity, funding details and conflict of interest) were added. The flowchart received positive feedback along with suggestions for several modifications from the PFGM.

Post-meeting activities

Based on the comments received at the face-to-face meeting, the steering committee modified and finalised the PRICE 2020 checklist and flowchart. The PRICE checklist and flowchart were then piloted by several authors to ensure they could be used during the development of real case reports.

The final PRICE 2020 checklist (Table 1) contains 12 sections/topics with 47 items (Title, Keywords, Abstract, Introduction, Informed and valid consent, Patient information, Discussion, Patient perspective, Conclusions, Funding details, Conflict of interest, and Quality of images used in the case report). Figure 1 is the PRICE 2020 flowchart consisting of 19 steps that summarise the sequence of stages involved when developing case reports.

Discussion

Case reports are considered an important part of the healthcare literature (Nissen & Wynn 2014). CAse REport (CARE) guidelines (Gagnier *et al.* 2013) were developed to provide guidance for authors when composing and critically assessing case reports in Medicine. The current project aimed to develop guidelines, exclusively for reporting cases in the field of Endodontics. The PRICE 2020 guidelines consist of a checklist with 12 sections and 47 individual items and a flowchart that should be considered when drafting an endodontic case report for publication.

Each section/topic of the PRICE 2020 checklist addresses individual components within a case report, with several items within each section. For example, the Title section has two items and the Keywords section has one; the Abstract has five, and so on (Table 1). In general, the number of items in each section reflects the importance (or weight) associated with a particular section of a case report.

Figures/images are an effective way of illustrating case reports and must, therefore, be self-explanatory (Kotz *et al.* 2013). Figures/images also provide evidence to support the text, the reported discovery and may help generate new research hypotheses (Kotz *et al.* 2013, Polepalli Ramesh *et al.* 2015); they are an important knowledge resource for biomedical researchers (Lang *et al.* 2012, Polepalli Ramesh *et al.* 2015). Given their significance, several items addressing the quality of figures/images in case reports were included in the checklist. This includes all images, e.g. radiographs, CBCT/CT/MRI scans, histology slides, clinical photographs

etc. To ensure all figures/images are of the highest quality and are useful, authors of case reports should consider all the nine items related to images.

Similarly, a pictorial representation in the form of a flowchart helps readers gain, at a glance, an overall view of the steps involved in developing case reports. Reporting guidelines with flow diagrams have also been developed to improve the quality of randomised controlled trials and systematic reviews, and to enable readers to understand the research process (Egger *et al.* 2001, Vu-Ngoc *et al.* 2018). Therefore, a flowchart was included as a component in the PRICE 2020 guidelines to emphasise the need to report on demographic details, patient concerns/symptoms, informed consent for investigation, medical history, dental history, clinical findings, diagnostic tests performed and their results, differential diagnosis, definitive diagnosis, management options considered, informed consent for treatment, treatment/interventions performed (if any), follow-up period(s), follow-up assessment method(s), treatment outcome, patient perspective and conclusion(s), funding details and conflict of interest.

Future plans

- 1. *Explanation and elaboration documents*: The steering committee will develop a detailed explanation and elaboration document to outline the rationale for each item in the PRICE 2020 guidelines. This will provide evidence and suitable examples from published case reports or hypothetical situations.
- 2. Translation: The PRICE 2020 guidelines will be translated into various languages.
- 3. *Dedicated website*: The PRICE 2020 guidelines (checklist and flow chart) published in the *International Endodontic Journal* will be linked to a new dedicated website: Preferred Reporting Items for study Designs in Endodontology (PRIDE) (www.pride-endodonticguidelines.org). Academics, researchers, journal editors, clinicians and students will be able to provide feedback on the guidelines via the PRIDE website, which will assist the steering committee when they are revised over time.

4. *Endorsement:* The project leaders will contact the Editors of relevant Endodontology and other dental journals to seek their support in adopting the PRICE 2020 guidelines, by adding the website link for the guidelines in their "Instructions to authors" or "Author information" or "Author guidelines" sections.

Conclusion

The PRICE 2020 guidelines have been developed by building global consensus for a checklist of items and a flowchart that can be used when composing case reports. The PRICE 2020 guidelines will help authors to prepare high-quality case reports in the field of Endodontics that will benefit all relevant stakeholders, including patients.

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Legends

Table 1: PRICE 2020 checklist of items to be included when reporting case reports in Endodontics.

Figure 1: PRICE 2020 Flowchart.

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Figure 1: PRICE 2020 Flowchart.

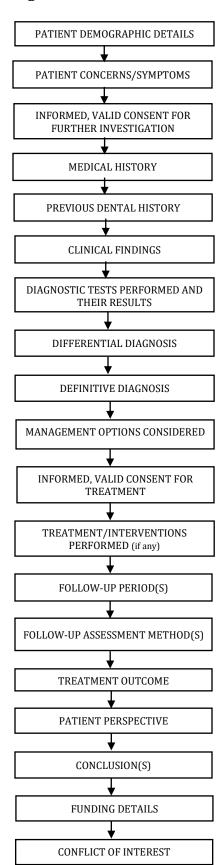


Table 1: PRICE 2020 checklist of items to be included when reporting case reports in Endodontics.

Section/Topic	Item number	Checklist Item	Reported on page number
Title	1a	The words "case report(s)" must be included in the title	
	1b	The area of interest (e.g. anatomy, disease, treatment) must be included briefly in the title	
Keywords	2a	At least two relevant keywords, preferably MeSH terms, related to the content of the case report must be included	
Abstract	3a	The Introduction must contain information on how the report is novel and contributes to the literature, clinical practice and/or fills a gap(s) in knowledge	
	3b	The Body must describe the main clinical findings, including symptoms and signs, if present	
	3c	The Body must describe the main radiographic/histological/laboratory/diagnostic findings	
	3d	The Body must describe the main outcomes of treatment, if active treatment has been provided	
	3e	The Conclusion(s) must contain the main "take-away" lesson(s), sometimes referred to as key learning point(s)	
Introduction	4a	A background summary of the case(s) with relevant information must be provided	
Informed consent	5a	A clear statement that informed, valid consent was obtained from the patient(s) must be provided	
Case report	6a	The age of the patient(s) must be provided	
information	6b	The gender of the patient(s) must be provided	
	6c	The ethnicity of the patient(s) must be provided, if relevant	
	6d	The main concern, chief complaint or symptoms of the patient(s), if any, must be provided	
	6e	The medical history of the patient(s) must be provided, if relevant	
	6f	The dental history of the patient(s) must be provided, if relevant	
	6g	The family history of the patient if associated with the primary complaint must be provided, if relevant	
	6h	The psychosocial history of the patient if associated with the primary complaint must be provided, if relevant	

	6i	Genetic information, including details of relevant comorbidities and past interventions and their outcomes must be provided when possible, if relevant	
	6j	Extra-oral findings must be provided, if relevant	
	6k	General intra-oral findings must be provided when relevant, e.g. carious lesions, restorations, periodontal condition, soft tissues etc.	
	61	Important/relevant dates and times (in the text, or a table or figure) must be provided in chronological order	
	6m	The diagnostic methods and the results for the specific tooth/teeth (e.g. pulp sensibility test, tenderness, mobility, periodontal probing depths, laboratory investigations, imaging techniques, or other special tests) must be provided	
	6n	The diagnostic challenges, if any, must be provided	
	60	The diagnostic reasoning including other possible diagnoses that were considered must be provided	
	6р	The active treatment (s) or intervention(s) performed, if any, must be provided	
	6q	Any modifications to the proposed treatment(s) or intervention(s), if necessary, must be provided	
	6r	The assessment method(s) used to determine the clinician- assessed and patient-assessed treatment outcomes and their results must be provided	
	6s	Adverse and unanticipated events or consequences, if any, must be provided	
Discussion	7a	The specific treatment(s) and intervention(s) (if any) must be discussed with reference to the relevant literature	
	7b	The strengths of the case report and its importance must be discussed with reference to the relevant literature	
	7c	The limitations of the case report must be discussed	
	7d	The rationale for the conclusion(s) must be discussed	
Patient perspective	8a	Feedback from the patient on the treatment and the care they received should be provided, if relevant	
Conclusion	9a	Explicit conclusion(s), i.e. the main "take-away" lessons must be provided	
	9b	Implications for clinical practice or future research must be provided	

Funding details	10a	Sources of funding and other support (such as supply of instruments, equipment) as well as the role of funders must be acknowledged and described
Conflict of interest	11a	An explicit statement on conflicts of interest must be provided
Quality of images	12a	Details of the equipment, software and settings used to acquire the image(s) must be described in the text or legend
	12b	The reason why the image(s) was acquired and the rationale for its inclusion in the manuscript must be provided in the text
	12c	The circumstances (conditions) under which the image(s) were viewed and evaluated by the authors must be provided in the text
	12d	The resolution and any magnification of the image(s) or any modifications/enhancements (e.g. adjustments for brightness, colour balance, or magnification, image smoothing, staining etc.) that were carried out must be described in the text or legend
	12e	Patient(s) identifiers (names, patient numbers) must be removed to ensure they are anonymised
	12f	An interpretation of the findings (meaning and implications) from the image (s) must be provided in the text
	12g	The legend associated with each image must describe clearly what the subject is and what specific feature(s) it illustrates. Legends associated with images of patients must describe the age, gender and ethnicity of the person, if relevant
	12h	Markers/labels must be used to identify the key information in the image(s) and be defined in the legend or as a footnote
	12i	The legend of each image must include an explanation whether it is pre-treatment, intra-treatment or post-treatment and, if relevant, how images over time were standardised