Case Report in Clinical Pediatric Dentistry: How to Review, Organize and Prepare a Manuscript

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A case report is a useful type of publication to describe information on unusual clinical diseases or syndromes, new management techniques, potential risk factors/oral disease associations, and uncommon side effects or responses to traditional dental treatments. In the practice of Dentistry and Medicine, the publication of case reports has the purpose of sharing new clinical experiences and knowledge with interested colleagues. Case reports in the field of Evidence-Based Pediatric Dentistry convey unique contributions to the clinical practice and help improve the process of clinical decision making in the form of a brief written communication. Additionally, case reports are potential resources of new hypotheses for more complex methodological designs in clinical research studies and are one of the best ways to get started in scholarly writing. The purposes of the present report were to comment on the role, relevance, and main limitations of case reports in Clinical Pediatric Dentistry, to describe the reasons for writing a case report and some recommendations for critically reviewing a published case report, and finally, to provide the fundamentals of preparing a case report manuscript in a structured manner.

Keywords: Pediatric Dentistry, Clinical case reports.

INTRODUCTION A case report is define

case report is defined as "a report of a single case of a disease, usually with an unexpected presentation, which typically describes the findings, clinical course, and prognosis of a case".1 In Clinical Pediatric Dentistry, the single case or case series reports are research documents that describe and analyze the diagnosis and/or oral management provided to one or more pediatric patients -including neonates, infants, children, and adolescents-.2,3 Case reports are considered as useful media to disseminate clinical information about uncommon or novel pediatric oral-related syndromes, risk/disease associations, and unusual side effects or responses to oral therapies. Sometimes, these papers contain a brief literature review of other similar previously reported cases, 1,4 - for example, see Miyashita et al.5 According to Friedman,6 despite that the case report has been less valued in the medical and dental literature, 250,000 case reports were listed in MEDLINE over a 5-year report period beginning in 1997, and many of the most important journals still publish case reports. Additionally, case reports are suitable tools for teaching the oral health sciences, and as learning experiences, to students, residents, or graduated dentists. Clinical case reports are also one of the best ways for authors to get started in scientific writing and to become familiarized with all aspects of publishing.^{2,7}

Clinical case reports often propose new theories or hypotheses that other clinicians may replicate in their own clinical practice, or present new information to guide the management of patients exposed to a same condition.³ In this context, the purposes of the

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present article were to comment on the role and relevance of case reports in Clinical Pediatric Dentistry, to describe the reasons for writing a case report, its main limitations, and some recommendations for critically reviewing critically a published case report. Finally, we attempt to provide the fundamentals for preparing a case report manuscript in a structured manner.

HOW TO WRITE A CASE REPORT IN CLINI-CAL PEDIATRIC DENTISTRY

Compared to other methodological designs or types of publication (e.g., randomized clinical trials), case reports lack specific sections such as *Materials and Methods*; also, they are not subjected to the thoroughness of blinded, hypothesis-driven clinical trials.^{7,8} The main reasons to write and publish a case report are the following:⁹

- To make an original contribution to the literature on a specific topic.
- To present unusual or understudied disorders, etiologic factors, clinical features, challenging differential diagnoses, or adverse effects with regard to oral care.
- To offer new insights into the pathogenesis of the oral disease.
- To describe improved or unique technical procedures in clinical practice.
- To describe possible errors in oral care, their causes and consequences.
- To present information that cannot be reproduced due to ethical reasons.
- To prompt a new hypothesis, to illustrate a clinical hypothesis, or to disconfirm previous stated hypotheses.

Additionally, the authors of a case report manuscript that intends to announce a new procedure or device, such as potential advances in the Clinical Pediatric Dentistry field, should carefully consider each of the following six questions related to the procedure or device before beginning to write the manuscript:¹⁰

- Is it needed? (Are current methods inadequate?)
- Is it logical? (Does it make good theoretical or physiological sense?)
- Does it work? (Has it been demonstrated that it actually does what it is supposed to do?)
- Is it safe? (Is there an added risk over standard methods and, if so, is the potential benefit worth it?)
- Is it an improvement? (Has it been shown to be better than what was available before?)
- Is it cost-effective? (Is the improvement likely to be worth the added time, effort, or expense?)

Types of case reports

According to Green *et al*,⁹ there are three types of case reports in the health sciences:

 (i) Diagnostic or assessment reports, to describe and discuss the diagnostic or analytic methods used to evaluate a pediatric patient with rare and confusing clinical features;

- (ii) Treatment reports, to describe the full management of a child, including an in-depth understanding of the case, and
- (iii) Educational reports, to provide current practice strategies together with a brief review of the literature. In general, there are two approaches for presenting a case report: the narrative or "storied" case report, and the evidence-based case report.^{9,11}

Regardless of the type of case report, it should include the diverse basic elements required to write a well-structured manuscript, in order to communicate the necessary and pertinent information for the reader.

Format

The traditional structural format includes:

- Title. According to Janicek, ¹² to be informative, and should include four elements: the intervention, the outcome, the population under study, and the condition. Let us see a good example: "Endodontic treatment of trauma-induced necrotic immature teeth using a tricalcium silicate-based bioactive cement. A report of 3 cases with 24-month follow-up". ¹³ In this title, the intervention is endodontic treatment using a tricalcium silicate-based bioactive cement. the outcome is the results after 24-month follow-up, the population- under-study is a pediatric patient with immature teeth, and the condition is necrotic immature teeth. A title such as this enables the reader to rapidly identify the topic presented.9
- Abstract. This is a brief presentation and includes the highlights of the reported case or cases. There are two types of Abstracts: narrative, and structured (with subsections comprising Introduction or Background, Case Report, and Conclusions). The majority of Clinical Pediatric Dentistry Journals call for Abstracts of a case report or of a case series that are limited to 150-200 words. Good examples of narrative and structured Abstracts can be found in Scheidt et al 14 and Mowafy et al,15 respectively. Immediately after the Abstract, it is necessary to write down 3-5 keywords, which can be used for describing the content of the case report and enable the full text of the article to be searchable online. They allow the reader to distinguish the most important ideas and concepts in the report. Each keyword should be kept short, one word where possible, though two and three word specialist terms are also acceptable where necessary.
- Introduction. It should include a brief but sufficient
 background on the topic, through an exhaustive review of
 the literature, to relate the context of the case in relation
 to previous published information; also, the introduction
 includes definitions of unusual terms and the purpose of the
 paper.¹¹ As an example, we recommend seeing Hong et al.¹⁶
- Case report (Methods and Results). Authors must mention
 the child assessment and clinical examination findings, as
 follows: 11 patient's complaint; medical/dental/social/family
 history; extraoral information (body, neck, skull, face, and
 lips, and temporomandibular joint); intraoral information

(caries, gingivo/periodontal and occlusal status; specific anomalies, hygiene level); x-rays taken and their justification; proposed diagnosis and differential diagnosis; and alternative and definitive treatment plans (prevention, pulp treatments, tooth restoration/rehabilitation, surgery, space maintainers/corrective orthodontics, periodontics, etc.), and also the prognosis, the case post-treatment evolution, and the oral health monitoring strategy. With respect to this, we can find two examples with a well-structured Case Report section, which are Ubaldini et al 17 and Hariri et al 18,

- **Discussion** (clinical implications) and **Conclusions.**^{9,11} Here, the authors manifest their opinions, and explain and discuss the case reported regarding known information and other published cases. Thus, several aspects should be mentioned in the Discussion section as follows: the differences between the treatment provided compared to that of the traditional practice; the manner in which the diagnosis was confirmed and the differential diagnosis; support of referenced drugs and materials; rationale for the management or the reasons for selecting one procedure over another; strengths and limitations of the case management and their significance; possible explanations for the final outcome -for example, unknown variables-, suggestions and hypotheses concerning the care provided or the results observed and why the care provided may or may not have been beneficial; practical recommendations, and recommendations for potential future research into the topic. We recommend Dangore-Khasbage et al 19 and Costa-Nogueira et al 20 and seeking out a good Discussion section.
- Complements: Ethical statement; acknowledgments; references; tables, figures (drawings or pictures), and their corresponding legends.

In terms of manuscript preparation, it is strongly suggested that researchers consult and follow in detail the "Instructions for Authors" established for the chosen journal, which are posted on its Website. These guidelines describe how to prepare, format, and submit a case report manuscript to increase its chance of acceptance. In the same context, Green et al.9 provide a pre-submission check sheet to assist in the writing process. Therefore, the presentation of a case report should be objective, avoiding extravagant claims or far- reaching conclusions. Authors should remember that the intention of their article is to describe and discuss a clinical event, not to prove anything, for example, a cause-effect relationship or the generalization of their findings to other, similar patients. 9,21,22

Common mistakes

According to Heller et al 7 and Pierson,10 a case report manuscript should be presented in a manner that convinces the Editor and Reviewers of its merits; thus, the main cause of rejection of a manuscript is no new relevant information about the case. Then, not following carefully the specific set of instructions and guidelines provided by every journal. These authors have mentioned the following 10 most common pitfalls encountered during the peer-review process as related to case report submission: (i) inexperience, (ii) inappropriate manuscript; (iii) insufficient information; (iv) incorrect format; (v) tunnel vision; (vi) incomplete or incorrect authorship information; (vii) improperly processed figures; (viii) improperly formatted references; (ix) suboptimal writing, and (x) failure to adequately revise a manuscript after peer review. Therefore, authors should pay attention to these details in order to facilitate the timely acceptance of their manuscript.

Ethical concerns

There are three crucial aspects to be considered when reporting a case in Clinical Pediatric Dentistry.9-11 First, it is mandatory to preserve the patient's identity, particularly the written identifying information and exhibited photographs. Second, permission should be obtained from the child's parents signed informed consent to publish the case; some journals require that the patient her/himself sign a specific form provided by the journal.9 And third, when the authors of a case report work for a university or hospital, and prior to submitting the report to a journal, they must send the manuscript to an Institutional Ethical Board for critical review and approval of its contents.10

How to Critically Review a Case Report in Clinical **Pediatric Dentistry**

Currently, there are available several checklists or guidelines to help authors of Clinical Pediatric Dentistry case reports to self-evaluate their manuscripts before submission to a journal, or for readers who want to evaluate the quality of a published case report. For example, Pierson¹⁰ provides a list of component domains to be rated when a reader wishes to evaluate the case report. In Pierson's scheme, there are five domains as follows: documentation; uniqueness; educational value; objectivity, and interpretation. Each domain is rated as follows: 2 (good); 1 (fair), and 0 (poor). After adding together all of the values, a total score is obtained and interpreted: 0-5: report is of insufficient quality for publication; 6-8: reader should be cautious about the validity and clinical value of the report; 9-10: report is a worthwhile contribution to the literature of Pediatric Dentistry.2

Additionally, in 2013, an international group of experts addressed the issue of standardizing case reports and developed the CARE checklist or guidelines (the term "CARE" is an acronym created from "CA", CAse, and "RE", REports), in order to increase the accuracy, transparency, and usefulness of case reports in the health sciences. This checklist provides a framework structured to evaluate the key components of a case report -grouped in 14 items-, and to capture useful clinical information.²³

Therefore, at present, there are different tools available for Clinical Pediatric Dentistry students, clinicians, and researchers in order for them to evaluate their own manuscripts or the evidence provided by previous published case reports or case series. 1 Through the critical reading of these works, it is wise to avoid applying, in our own pediatric patients or investigations, evidence deriving from ill-advised, dangerous, or expensive devices or procedures that appear in the literature simply because they are new and different. 8,10

LIMITATIONS

Readers of case reports in Clinical Pediatric Dentistry should consider that this type of publication has limitations. First, the management strategy of a specific patient or group of patients occurs primarily in an uncontrolled environment, in which the effect of confounding variables -for example, the level of compliance of the reported child-cannot be avoided; in other words, the reported patient response may not be due exclusively to the treatment provided. Second, because the management rendered to one patient may not produce the same effects in another patient with a similar condition, the informed results or findings cannot be generalized beyond the particular context of the reported patient. Third, the results of a case report (or case series) may have been influenced by the natural history of the patient's disease. Some oral conditions may exhibit spontaneous remission or stages of remission/exacerbation. It is possible that one of these stages corresponds to the time that treatment was administered, or, contrariwise, the child could begin her/his care immediately prior to the presentation the worst part of a disease. In both situations, biased conclusions could be generated. Finally, many case reports are based on available retrospective data from incomplete or deficient medical or oral health records, introducing a potential error in the report.^{2,9,11}

FINAL COMMENTS AND CONCLUSIONS

Historically, case reports have been considered an honored tradition in the medical and dental profession.²² Published case reports have provided rich information for optimal patient care, including relevant scientific observations or individual clinical insights that may be missed or undetected in other, more sophisticated research designs.^{1,11,22}

The National Health and Medical Research Council (NHMRC) considers that case reports belong to level four of the evidence hierarchy for adequate clinical decision making process in pediatric oral health, because they are much less convincing than other methodological designs, such as systematic reviews, randomized clinical trials, and cohort or case-control studies.^{3,8} In the era of Evidence-Based Pediatric Dentistry (EBPD), well-documented case reports constitute an invaluable resource for providing preliminary evidence and a useful guide for future, larger-scale clinical research with increased quality.^{8,9}

As mentioned previously, many prestigious medical and dental journals continue to consider and publish case reports, to benefit of the clinical practice worldwide. The Clinical Pediatric Dentistry field is not an exception. In a literature search in PubMed, we found a total of 857 references on clinical case reports published during the period from 2007–2017. The search algorithm was as follows: ("paediatric dentistry" [All Fields] OR "pediatric dentistry" [MeSH Terms] OR ("pediatric" [All Fields] AND "dentistry" [All Fields]) OR "pediatric dentistry" [All Fields]) AND (Case Reports[ptyp] AND "2007/06/17" [PDat]: "2017/06/13" [PDat]). Thus, although the case report or case series does not possess as much impact on clinical research as a systematic review or a randomized clinical trial, this type of publication is still essential and necessary for documenting novel or rare syndromes, manifestations, associations, outcomes, or complications. When properly written, case reports possess the potential to aid readers to better understand the oral care of individual child patients and may lead to advances in research, thus contributing to the improvement of the Clinical Pediatric Dentistry practice worldwide.

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