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Traumatic dental injury research: *on* children or *with* children?

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Conflict of interest

All authors declare no conflict of interest.

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Abstract

Background and aim: It is widely acknowledged that children should participate in healthcare decisions, service development and even setting research agendas. Dental traumatology is a major component of paediatric dentistry practice and research. However, little is known about young patients' contribution to new knowledge in this field. The aim of the study was to establish the extent to which children are involved in contemporary dental trauma research and to evaluate the quality of the related literature.

Material and methods: A systematic review of the dental trauma literature was conducted from 2006-2014. The electronic databases, MEDLINE and Scopus, were used to identify relevant studies. The selected papers were independently examined by five calibrated reviewers. Studies were categorised by the degree of children's involvement and appraised using a validated quality assessment tool.

Results: The initial search yielded 4,374 papers. After application of the inclusion and exclusion criteria only 96 studies remained. Research *on* children accounted for 87.5% of papers and a proxy was involved in 4.2%. Children were engaged to some degree in only 8.3% of studies and there were no studies where children were active research participants. In the quality assessment exercise papers scored, on average, 57% (range=14-86%).

Conclusion: There is scope to encourage more active participation of children in dental trauma research in the future. Furthermore, there are some areas where the quality of research could be improved overall.

Introduction

Over the past decade, there has been increased focus on the need for patient and public involvement in health care planning and development (1). Patients have a right to voice their views in order to improve services. There has been a similar drive to involve patients more meaningfully in health research, with many funding bodies stipulating the need for patient input within the application process. A review assessing the effect of patient and public involvement on health and social care research found that public involvement increased recruitment rates, improved the quality of data and facilitated dissemination of the findings (2). These fundamental changes in how patients are viewed and engaged are not limited to adults. Children's opinions about what matters to them are also being more widely sought by health professionals (3, 4). Children can be involved at different stages in the research process, from proposing research priorities, shaping the design and development of the study, conducting the fieldwork, through to analysis and dissemination of the findings. A variety of child-centred quantitative and qualitative approaches have been described, each with its own strengths and limitations (5, 6). The use of oral health-related quality of life (OHRQoL) questionnaires is a rapidly expanding field and a number of these measures have been developed specifically for children (7-10).

A landmark systematic review of the paediatric dental literature, published from 2000 to 2005, was undertaken to assess the extent of children's involvement in oral health-related research (11). The authors found that only 7.3% of studies had been carried out *with* children as active participants. Recommendations were made and opportunities highlighted to promote greater involvement of children in future oral health research. It was a welcome finding therefore, that in a subsequent review of

the contemporary paediatric dental literature, published between 2006 to 2014, the proportion of studies conducted *with* children had increased to 17.4% (12). These systematic reviews were the first to attempt to quantify the involvement of children in oral health-related research, and they identified the need and scope for more active participation of children.

Dental traumatology constitutes a major component of paediatric dentistry practice and research. As new materials and techniques emerge, as well as a greater understanding of the impacts of dental conditions and related treatments, it is important that researchers reappraise patients' views and opinions in relation to traumatic dental injuries (TDI). Such enquiry will help identify what is important to young patients and to prioritise where improvements can be made to better meet their needs. With this in mind, an appraisal of the paediatric dental trauma literature was felt to be warranted.

The aim of this study was to undertake a systematic review of the contemporary dental trauma literature published from 2006-2014 to determine the degree of children's involvement in clinical research. The specific objectives were to:

1. determine the context of paediatric TDI-related clinical research with regards to country of origin, setting and study design;
2. categorise contemporary dental trauma research by the degree of children's involvement; and
3. assess the quality of the literature using a validated quality assessment tool.

Material and methods

Search strategy

An electronic search was conducted using keywords on databases MEDLINE (via Ovid) and SCOPUS from 1st January, 2006 to 28th March, 2014.

The keywords used were:

- child*
- AND
- dental OR tooth
- AND
- avulsion OR concussion OR subluxation OR extrusion OR intrusion OR luxation OR injuries OR trauma OR fractures

The following limits were applied: human studies; English language; publication year 2006-2014; 0-18 year olds and dentistry journals. Our inclusion criteria specified participants were to be 16 years and younger, however the most suitable age limit option on the databases was 0-18 year olds.

Selection criteria

References identified by the search were exported to Endnote and duplicates were removed. The first stage involved two reviewers who independently applied the exclusion criteria to titles and abstracts. Where there was insufficient information detailed in the abstract, the full paper was reviewed at the next stage. Any disagreement was resolved through discussion.

The following exclusion criteria were applied to the search:

- studies published prior to 2006: a systematic review assessing the extent of children's involvement in child dental literature had been previously conducted from 2000 to 2005 (11), therefore the optimal timeframe to include in the present review was from 2006 to 2014
- studies with participants over 16 years of age
- languages other than English
- non-dental journals
- studies that did not have children and TDI as their main focus
- retrospective case note review only
- *In vitro* studies
- studies with no primary data
- conference proceedings

The following inclusion criteria were applied to the search:

- timeframe (2006-2014)
- human studies
- written in the English language
- participants 16 years of age and younger

Data collection

The accepted papers were examined and data were extrapolated relating to the following categories:

- year of publication
- title of journal

- study design (cross-sectional, longitudinal, or observational)
- country where the study was conducted
- study setting (pre-school/school or dental clinic)
- number and age of participants
- dentition(s) studied (primary, permanent or both dentitions)
- category of children's involvement (detailed below)

The included studies were classified by the level of children's involvement using a categorisation framework first developed by Marshman et al. (11) (Table 1). This system broadly ranked children's participation into one of four categories:

1. research conducted *with* children as active participants;
2. research conducted with children but where the children took the role of subject;
3. use of a proxy on behalf of children;
4. research conducted *on* children as objects of the research enquiry.

The four main categories were further defined into seven subgroups.

To achieve the third objective, the papers were appraised using a validated 16-item quality assessment tool, known as QATSDD (13). This instrument is a unique quality assessment tool, developed at the University of Leeds, UK, which can be applied to diverse study designs, unlike the majority of other tools which evaluate a single methodological approach. Each item was scored according to the degree to which each quality criteria was met: 0=not met at all; 1=very slightly met; 2=moderately met and 3=completely met. Of the 16-items, two items (criteria 9 and 10) are specific for quantitative studies and a further two items (criteria 11 and 14) are specific for qualitative studies. Thus the minimum score that could be awarded to a paper

employing a single method would be 0 and the maximum would be 42. In the case of a mixed methods study design, the maximum achievable score would be 48. The quality criteria relate to the following broad categories:

1. Explicit theoretical framework
2. Statement of aims/objectives in the main body of the report
3. Clear description of research setting
4. Evidence of sample size considered in terms of analysis
5. Representative sample of target group of a reasonable size
6. Description of procedure for data collection
7. Rationale for choice of data collection tool(s)
8. Detailed recruitment data
9. Statistical assessment of reliability and validity of measurement tool(s)
(quantitative only)
10. Fit between stated research question and method of data collection
(quantitative only)
11. Fit between stated research question and format and content of data collection tool e.g. interview schedule (qualitative only)
12. Fit between research question and method of analysis
13. Good justification for analytical method selected
14. Assessment of reliability of analytical process (qualitative only)
15. Evidence of user involvement in design
16. Strengths and limitations critically discussed

Five reviewers took part in the systematic review following a training exercise to ensure consistency of data extraction, categorisation of children's involvement and application of the quality assessment tool. The data collection form was piloted using three papers. Minor adjustments were made to the form as necessary after discussion between the reviewers. An initial calibration exercise was conducted using five papers. The level of agreement between examiners was calculated simply as 'yes' or 'no' for all categories and the number of instances where there was agreement was calculated as a percentage of the total number of items where a decision was recorded.

The full texts of the papers that met the abstract screening criteria (n=222) were retrieved and each pair reviewed a third of the papers independently of each other. Papers which matched the exclusion criteria were rejected. The accepted papers were then examined (n=96), relevant data extracted and the quality appraised. The reviewers met in their pairs to resolve any disagreements. After an 8-week interval, 10% of the accepted papers were re-examined to calculate both inter-examiner and intra-examiner agreement.

Results

The initial search strategy yielded 4,374 articles, as shown in Figure 1. These were exported to Endnote X7 and duplicates removed, resulting in 3,504 papers. After application of the exclusion criteria, 222 studies remained. Once the complete texts were examined, 126 articles were found not to meet the inclusion criteria and were rejected, leaving 96 papers for analysis.

Journal

Over half of the papers (57.3%, n=55) were published in *Dental Traumatology*. The remaining 42.7% (n=41) of articles were published across 22 different journals. Seven of these were specific paediatric dentistry journals and these collectively accounted for 17.7% (n=17) of the publications under review.

Country of study origin

The 96 studies had been conducted in 26 different countries, which included a combination of developed and developing countries. However, the majority of the dental trauma papers were attributed to work undertaken in Brazil (38.5%, n=37), followed by India (12.5%, n=12) and Turkey (7.3%, n=7).

Study design

Most of the studies were cross-sectional in design (75.0%, n=72). Longitudinal observational studies and randomised controlled trials accounted for only 14.6% (n=14) and 2.1% (n=2) of the papers respectively. No qualitative studies were identified for inclusion in this review.

Setting

Just over half of the studies (57.3%, n=55) were conducted in a pre-school or school environment and a quarter (28.1%, n=27) recruited participants in a dental clinic setting. The remainder were conducted in general hospitals and other locations, which each accounted for 7.3% (n=7) of the studies.

Dentition involved

The permanent dentition was the focus of 60.4% (n=58) of the papers and less than a third (30.2%, n=29) of the articles related to a TDI involving the primary dentition. A small number of studies (6.3%, n=6) looked at both dentitions and in 3.1% (n=3) of the studies, the dentition was not specified.

Involvement of children

Research was categorised as being *on* children (Category 4) in 87.5% (n=84) of papers and a parent proxy (Category 3) was involved in 4.2% (n=4) of cases. Of the studies which included a parent proxy, one had used a quality assurance questionnaire (14) and another had involved a questionnaire about scooter-related accidents (15). Two papers had measured the parents' perception of the impact of TDI on the child's quality of life using the Parental-Caregiver Perception Questionnaire (P-CPQ) for 8- to 14-year-olds (16) and the Early Childhood Oral Health Impact Scale (ECOHIS) for 5-year-olds (17).

Only 8.3% (n=8) of the studies engaged children to some degree (Category 2) and no study was deemed to fully involve children as active participants (Category 1). A questionnaire was the sole approach used to seek children's perspectives. Of these enquiries, three had assessed patient satisfaction with appearance (18-20), one article looked at children's experiences and knowledge of dental trauma (21), and four papers measured the impact of dental trauma on quality of life (22-25). The CPQ₁₁₋₁₄ was the sole instrument used to evaluate child-reported OHRQoL.

Research quality

On average, the papers scored 56.7% (equating to 23.8 marks out of a maximum of 42) in the quality assessment exercise (SD=18.7%; range=14.3-85.7%). Areas which performed the best related to: description of data collection procedure; suitability of the method of data collection, and appropriateness of analysis to answer the research question. Poorest performance related to: a lack of an explicit theoretical framework; absence of a sample size calculation; poor user involvement, and sparse discussion of strengths and limitations (Table 2).

Inter-examiner and intra-examiner agreement

Inter-examiner agreement for general data extraction and categorisation of child involvement was excellent (range=84-97%) but was slightly poorer for agreement with respect to the quality assessment scores (54-82% agreement). Intra-examiner agreement, which was determined after an 8-week interval, was good for both the general data extraction as well as the quality assessment scores (92-98% and 64-76% respectively).

Discussion

It was not surprising to discover that *Dental Traumatology* had published the majority of articles included in this review, since, it is the only journal to have TDI as its sole focus. Furthermore, it has a relatively high impact factor (1.601), indicating access to a large audience and making it more popular with prospective authors. However, the reason for the high proportion of publications emanating from Brazil (over one third of all included studies) warrants further consideration. One explanation may be that Brazil has a large number of dental schools training high numbers of dentists and

potential researchers each year. Indeed, over the past decade there has been major expansion in dental education, with Brazil now exceeding the United States and all of Europe in the number of dental schools and students (26). It is also evident, from the wider literature, that Brazilian dental academics have had a longstanding interest in OHRQoL research.

Disappointingly, there were no qualitative or mixed method studies found in this literature review. It is acknowledged that conclusions drawn from systematic reviews may be obsolete almost as soon as they are published, due to the rapidly growing literature base (27). The need for regular updating is therefore recognised, and with this in mind, a further search of the most recent TDI literature (until April 2016) was undertaken. This again failed to identify any qualitative studies involving children who had sustained a TDI. A reason for this may be that dental researchers require specific training or input from social science experts to conduct these studies which may be a barrier to some. Furthermore, researchers may not yet fully appreciate the value of undertaking qualitative research within the field of paediatric dental trauma. There is also a common perception that the scientific rigour of qualitative studies may not match those with a quantitative methodology, which may account for the lower acceptance rates of such papers by many journals. Conversely, some would argue that without appropriately conducted qualitative enquiries, opportunities are being missed to gain meaningful insights into the child's perspective of TDI.

The main focus of this study, however, was to determine children's engagement in TDI-related research. Interestingly, a parent proxy was involved in relatively few (4.2%) papers (14-17). The use of proxies may have a place in oral health research

but remains a controversial subject. Jokovic and colleagues (28) investigated the agreement between parental and child reports using the Parental Perception Questionnaire and the CPQ₁₁₋₁₄. They found that some parents had a limited understanding of their child's OHRQoL, particularly with respect to impacts on emotional and social well-being. It is speculated that, as these impacts may relate to activities outside the home environment, parents may not always witness them. Furthermore, this apparent lack of parental insight was more pronounced for older children (11-14 years old), who may spend increasing time away from their parents, and thus share fewer experiences together.

It is worth noting that of the four studies that did rely on proxy reporting, three involved children aged 8-years and older. Therefore the use of a self-report questionnaire may have been more appropriate. With certain groups it may prove difficult for researchers to involve children directly, such as those under the age of five and children with profound learning difficulties. However, a number of communication barriers can be overcome with appropriate approaches (29). It is suggested that parental reports may be used to complement child reports rather than substitute for them.

In this review, only a relatively small proportion of studies (8.3%, n=8) were found to have engaged children to some degree. Of these, four measured the impact of TDI on quality of life using the CPQ₁₁₋₁₄, with three using a short version of the same instrument (30). As with the P-CPQ, the CPQ was validated using participants primarily with caries, malocclusions or cleft lip and/or palate (7). Therefore, findings must be interpreted with some caution when these generic OHRQoL measures are

used for children and young people with specific conditions, such as TDI, as they may not be sensitive enough to identify a correlation between the dental condition and the resultant numerical scores.

There were no investigations which were considered to fully engage children in the study design and conduct. Gilchrist and colleagues (6) discussed a number of approaches which can be utilised to involve children in oral health research, such as interviews, focus groups, questionnaires and diaries. Involving children in research provides several advantages - for example, children can help identify the research question, ensuring that subsequent investigations produce outcomes that are relevant to them. They can also contribute to the development of resources, such as patient information leaflets or reports, to ensure they are in an understandable and engaging format. Research that focuses purely on clinical outcomes following a treatment intervention and fails to consider the patient's experience of that intervention may be open to question in terms of its overall value and relevance.

The third objective of this review was to appraise the quality of the included TDI literature. No authors referred to an explicit theoretical framework or model to underpin their work. This is a difficult concept and may not have been relevant to all study designs. Indeed the developers of the QATSDD (13) acknowledge that some of quality criteria may not be suitable for certain study designs. Clearly, epidemiological studies do not necessitate an underpinning theoretical model, nor do studies which have evaluated clinical outcomes. Theoretical frameworks may be more appropriate to studies with a qualitative component and they may help to inform the study design and explain findings. The absence of any qualitative studies

therefore makes the results less remarkable. It is acknowledged that OHRQoL measures, such as the CPQ, have been driven by an underlying theoretical framework but if this was not stated explicitly in the paper, a score of zero was attributed, according to the QATSDD criteria. Interestingly, a study published after this systematic review had been conducted did propose a theoretical framework of resiliency and adaptation in order to explore children's outcomes following a TDI (31).

Other areas which were also poorly addressed in the studies are less defensible - such as inclusion of a sample size calculation, user involvement, and discussion of strengths and limitations. It is important that authors appraise the strengths and limitations of their work so that the conclusions are accurately interpreted and solutions can be suggested for any future research. An adequate sample size is essential to provide a study with sufficient statistical power so that any statistically significant differences can be identified, if they exist. This methodological consideration is becoming an essential part of journals' acceptance criteria for published studies and should therefore drive future improvement in this area.

Few studies incorporated any user involvement in their study design, which is disappointing. As mentioned earlier, Staley (2) found that public involvement in the research process brought many benefits, and where possible, attempts should be made to engage children and young people throughout the research process. It is hoped that improvement will be seen in this area as funding bodies increasingly demand that participants are actively involved in grant applications and research

protocols. Health research must strive to address issues that patients feel are important rather than just those that clinicians believe are a priority.

It is worth noting that the standard deviation for the scores ascribed to studies in the quality assessment exercise was high (18.7%). One explanation for this may be a true marked difference in quality of the papers. Characteristics of the QATSDD, rather than the study quality per se may explain the low scores for some papers, and hence the high standard deviation.

The QATSDD uses a 4-point scoring system to try and provide a more accurate representation of the paper's quality. However, this scaled scoring system means there is a wider margin for disagreement between reviewers compared to a 2-point scoring scale. The authors acknowledge this limitation and advise reviewers to independently appraise the papers, and following this they should meet to discuss and resolve any differences. This process was therefore integrated into the present systematic review's methodology.

Guidance notes are provided alongside the QATSDD scoring system. These notes, together with the reviewer's judgement and knowledge in research methods, are used to critically appraise the study. The research experience of the reviewers in this present review was quite varied and may account for the inconsistencies in inter- and intra-examiner agreement. Furthermore, it was felt that the QATSDD would benefit from the provision of greater detail in some of the descriptions to improve inter-examiner agreement. However, the QATSDD was found to be generally applicable to the range of study designs encountered in this review. Unfortunately,

no comment can be made on the use of the tool for qualitative research as no such studies were identified.

The inclusion criteria for this systematic review specified the study must be available in English language. Studies from non-English speaking countries will have been rejected, therefore this limitation should be recognised when interpreting the findings. A further limitation of the review was the exclusion of studies using both child and adult participants due to the difficulties involved when trying to extrapolate and analyse data for the child participants only. It is therefore recognised that these papers may have provided additional data and insights into the degree to which children are involved in trauma-related research. Nonetheless, it is felt that the key findings identified by review are topical and informative for both clinicians and researchers even though the literature is continually evolving.

In summary, it is clear that there is considerable scope for clinical investigators to engage children more actively in trauma-related research, thereby ensuring that research enquiries address issues that are important to patients themselves. Furthermore, this review highlights some areas where the quality of future studies can be improved by adopting accepted good practice as outlined in quality assessment tools such as QATSDD (32).

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Category	Properties	Code
1. <i>With</i> children – children seen as active participants	Children involved in the research process	1
	Children’s own accounts incorporated	2
2. <i>With</i> children – children seen as subjects	Children completing measures designed by adults	3
	Case report/series with child’s input throughout case	4
3. Proxies for children used	Parent/caregiver used appropriately as proxy	5
	Clinician used appropriately as proxy	6
4. <i>On</i> children	Children seen as the objects of the research	7

Table 1: Categorisation framework of children's involvement according to Marshman et al. (11)

Quality criteria	Mean score (SD, range)
Explicit theoretical framework	0.00 (0, 0)
Statement of aims/objectives in the main body of the report	2.38 (0.8, 0-3)
Clear description of research setting	2.29 (0.7, 0-3)
Evidence of sample size considered in terms of analysis	1.24 (1.4, 0-3)
Representative sample of target group of a reasonable size	2.08 (0.9, 0-3)
Description of procedure for data collection	2.44 (0.7, 1-3)

Rationale for choice of data collection tool(s)	2.01 (1.0, 0-3)
Detailed recruitment data	1.81 (0.9, 0-3)
Statistical assessment of reliability and validity of measurement tool(s) (quantitative only)	1.54 (1.4, 0-3)
Fit between stated research question and method of data collection (quantitative only)	2.59 (0.6, 0-3)
Fit between stated research question and format and content of data collection tool e.g. interview schedule (qualitative only)	non-applicable
Fit between research question and method of analysis	2.48 (0.8, 0-3)
Good justification for analytical method selected	1.76 (0.9, 0-3)
Assessment of reliability of analytical process (qualitative only)	non-applicable
Evidence of user involvement in design	0.20 (0.5, 0-2)
Strengths and limitations critically discussed	1.01 (0.9, 0-3)

Table 2: Table showing mean score (SD, range) for each of the quality criteria cited in Sirriyeh et al. (13). Quality criteria: 0=not met at all, 1=very slightly met, 2=moderately met and 3=completely met.

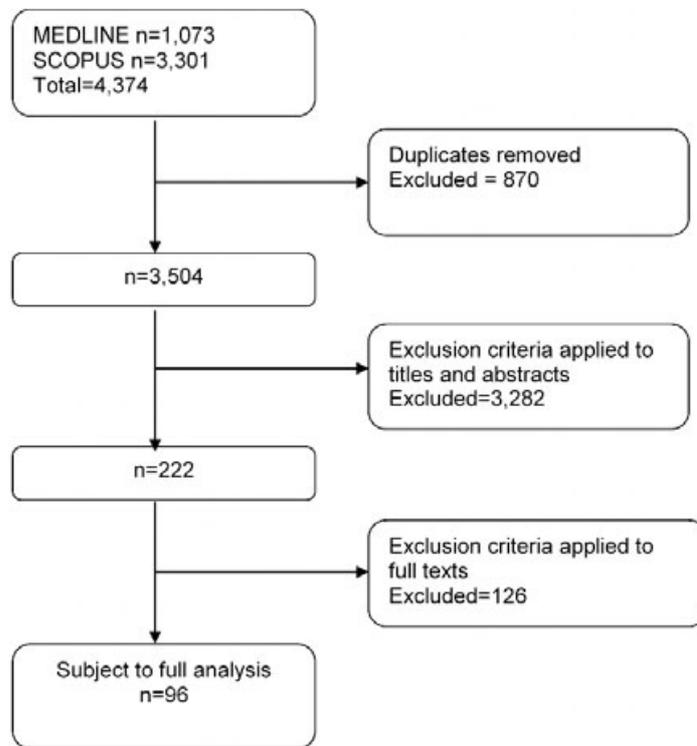


Figure 1: Flowchart detailing the review process for included studies