ORIGINAL ARTICLE

The inclusion of children with disabilities in oral health research: A systematic review

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Abstract
Objective: This systematic review aimed to describe the extent to which oral health research since the advent of the United Nations Convention on the Rights of the Child, the United Nations Convention on the Rights of Persons with Disabilities and the current movement on the inclusion of voices of children, has been done on or with children with disabilities.

Methods: A systematic review of the dental literature for children with disabilities from 2001 to 2017 was conducted. Electronic databases were searched for oral health literature on the involvement of children with disabilities. Each identified article was examined by 2 reviewers against the inclusion criteria and categorized according to the extent to which children with disabilities were involved in oral health research, type of study, children’s ages, type of impairment and the country of origin.

Results: The search included 113 articles after application of the exclusion criteria. Of these papers, 6.2% were classified as research with children, 41.6% used proxies to gain the perspective of children with disabilities and 52.2% were classified as research on children with disabilities. The majority of studies, which attempted some form of inclusion, were from Sweden and India.

Conclusions: Most oral health research is conducted on children with disabilities (viewing them as objects) rather than with them (viewing them as active participants). Unlike previous systematic reviews which report an increase in the inclusion of the voices of children in oral health research, children with disabilities were mostly excluded from research. Future research should attempt to use methods which involve children with disabilities as fully as possible in the research process. This ensures that their perspectives are obtained and their voices are heard.

KEYWORDS
children, dentistry, disabilities, oral health, inclusion

1 | INTRODUCTION

The United Nations Convention on the Rights of the Child (UNCRC) was held in 1989 to promote support for the rights of children. This resulted in children with disabilities gradually beginning to claim their rights, both as children and as disabled individuals. Of the UNCRC’s 54 articles, article 12 was arguably the most influential of the articles for research, because it argued that children should have the right to express their opinions and to have their perspectives taken seriously in matters that affect them, including health research. This was also significant for all children, because their rights were upheld regardless of race, family, colour, religion, language, politics, sex, nationality, social origin, ethnicity, disability, birth and so on; they were seen as a diverse body. This is evident in Article 2 of the UNCRC, which established the principle that all children’s rights should be promoted without
discrimination; therefore, the convention also has a profound impact on how children with disabilities are perceived.3

In the Western world, the majority of children with disabilities attend mainstream schools, live at home with their families and have equal rights to treatment and inclusion as children without disabilities which is enshrined in International Convention:

\[\text{[\ldots] shall ensure that children with disabilities have the right to express their views freely on all matters affecting them, their views being given due weight in accordance with their age and maturity, on an equal basis with other children, and to be provided with disability and age-appropriate assistance to realise that right. (The United Nations Convention on the Rights of Persons with Disabilities) [UNCRPD], 2009 Article 7}\]

Even though children with disabilities are children first, they are often left out from “generic children’s research, from policy-making about children’s services and, in their everyday lives, from inclusion in friendship groups and social and sporting activities” (p.173).2 Children with disabilities may need additional support to conduct their everyday lives but they have the same needs, hopes and wishes as any other young person.

In the field of childhood studies, the UNCRC has enhanced children’s level of involvement in research by valuing their right to have a voice and have their opinions heard.3 This research perspective views children as experts on their own lives who can provide valued knowledge to validate, enrich and sometimes challenge the evidence base.4 One aspect raised within the sociology of childhood is the tendency to omit social categories of significance such as ethnicity, social class and disability, which intersects with who they are; their social position and the necessity for researchers to incorporate these aspects into the research design thereby making research more appropriate and inclusive for children.5 This vision of the child as a diverse agent would appear important if research is to shift from portraying children in terms of a narrow range of social categories.6

The concept of the voices of children in health-related research appears to have emerged more slowly than in other fields, largely because of the entrenched protectionist and paternalistic perspectives that have historically pervaded disciplines such as medicine.7 Although, there may also be an element of safeguarding involved and this could also be a reason for the exclusion of children from research.8

With the emergence of the new sociology of childhood and disability studies, there has been a new approach to the study of disability in childhood with a shift from exploring impairment towards placing the child at the centre of the research process.5 Historically, research relied on adult perspectives to shape meaning rather than the views and experiences of children with disabilities9 or concentrated on children who are verbally articulate.10 Nevertheless, an increasing number of social researchers are focusing on using qualitative methodologies to determine the views of children with disabilities in research to improve understandings of what is important to them. These academics argue that it is not acceptable to exclude the voices of children with disabilities merely as they pose a challenge to traditional research approaches, for example if they lack verbal articulacy. Indeed, the responsibility is on academics to consider diversity more strongly when they design their research and develop skills to adopt inclusive research strategies that enable children with disabilities to become active participants in research that concerns them.11-15

Within the field of oral health research, 2 valuable systematic reviews have been conducted to show how far dental research has been carried out with or on children. The first systematic review found that the majority of research (87%) of papers viewed children as the “objects” on which to do research on, rather than with.16 Another systematic review of the dental literature from 1997 to 2007 involving adults with intellectual disabilities was also conducted by Whelan et al,17 using the 4 main categories of Marshman et al18 to synthesize the papers. Of the 173 papers reviewed, most of the published dental research (82%) involving adults with intellectual disabilities treated them as research objects. Only 2.2% of the studies actively involved them; 9.3% involved them to some extent; and 5.2% used proxies to represent the views of adults with intellectual disabilities.17 Marshman et al18 then carried out an updated systematic review which identified that the proportion of research on children had decreased by 23% over that decade and moved more towards research with children rather than on them. What neither child systematic review does is identify whether children with disabilities are viewed as a diverse group and if they are included in oral health research. The aim of this study was to conduct a systematic review of the extent to which contemporary oral health research has been conducted with or on children with disabilities.

2 | METHOD

This systematic review was based on the former reviews of oral health research involving children.16,18 This study adapted the categories that were developed by Marshman and colleagues (2007), with some modification based on the wider social science literature around the inclusion of children with disabilities in research and collected published articles about the oral health of children with disabilities from 2001 to 2017. These categories are described below.

1. The first category included research with children with disabilities where children were seen as active participants sharing power and responsibility for the research design and process. Children with disabilities are included in all aspects of the research process from generating the questions through to the design as well as being involved in data collection and dissemination. Children are viewed as equals.

2. The second category, also classified as research with children with disabilities, where children are involved in the design and conduct of research. Children worked within pre-arranged focus and research questions and were involved in choices of methods and ways of carrying out research.

3. The third category included research with children with disabilities where their perspectives were taken into consideration
around research design. The children would express their views around the research design and were involved in steering groups.

4. The fourth category also comprised research with children with disabilities and comprised studies where they were listened to and seen as subjects of research with adults deciding the research questions, design and methods. This category was subdivided into 2 subcategories. The first subcategory included studies where children with disabilities contributed by giving an account of their experiences in their own words (subcategory 4a), using methods for example qualitative interviews, in-depth, unstructured and semi-structured. The second subcategory-contained studies where children with disabilities completed measures designed by adults (subcategory 4b) for example, structured interviews, questionnaires and other scales.

5. The fifth category included studies that used others as proxies for children with disabilities—another person reporting on their oral health either parental/caregiver (category 5a) or clinician (category 5b). Using proxies was felt appropriate for children too young or sick or with a profound level of intellectual disability.

6. The final category included research on children with disabilities, where they were seen as the objects of research. In this research, children with disabilities were not consulted in any way, but only seen. For example, research included within this category might see them either as; a mouth or a set of teeth to be treated; source of a sample of plaque, saliva or soft/hard tissues; recipient of an intervention such as oral health promotion; population group to be examined clinically; particular group to be managed and finally a patient on whom a “particular examination” was done.

These key categories alongside their sub categories were adapted to classify the included studies in the current systematic review. The search strategy was undertaken to identify all published oral health studies on children with disabilities between 2001 and 2017 reflecting the conceptual model of the International Classification of Functioning, Disability and Health and “Valuing People” in the UK using child-related keywords, disability-related keywords and dental-related keywords as research terms. The full search strategy including MeSH terms is available in Appendix S1.

The search strategy was performed using Scopus, Web of Science (which incorporates 12 online databases, alongside Medline® and the social sciences databases used by the previous Web of Knowledge) and Google Scholar which was used to search for any peer-reviewed articles that may not be included in the databases. It was limited to dental journals and the English language. The resultant articles were exported to an Endnote library and duplicates were removed at this stage. The titles and abstracts of each reference were screened independently by 2 trained reviewers based on the following exclusion criteria; studies reported before 2001 because this was prior to the conceptual model of the International Classification of Functioning, Disability and Health and “Valuing People” in the UK; studies with participants over 16 years of age; studies that do not have children with disabilities and/or aspects of their oral health as their main topic; studies with no primary data, case reports, conference proceedings and guidance documents.

Agreements about the application of exclusion criteria between the 2 reviewers occurred for 86% of the articles, and disagreements were settled by discussion between the reviewers and referral to a third reviewer if agreement could not be reached. Full texts of papers that passed the initial title and abstract screening were retrieved to determine eligibility for inclusion in the review. At this stage, studies that did not meet the inclusion criteria were excluded and the reason for exclusion was noted.

Two researchers involved in the categorization classified 5 papers together as a training exercise and an additional 5 papers independently for calibration purposes. If categorizing the paper from the abstract was not possible, the full text of the article was retrieved. If the papers appeared to fit in more than one category, the category that assumed the greater involvement of children with disabilities was chosen. The agreement between the reviewers about categorization occurred for 97% of the papers, and disagreements were solved by discussion between the reviewers or by recourse to a third reviewer if this proved difficult. The frequency distribution of the categories of articles can be seen in Table S1.

Data were extracted by 2 reviewers independently, using an EXCEL spreadsheet to record the following: author name, publication year, type of study, the country where the study was conducted, sample size, the age of children and type of impairment. Discussion solved disagreements in data extraction and, if necessary, a third member of the review team was involved. Alongside the categorization of papers, the social model of disability was also used as a lens through which to review and synthesize the papers.

3 RESULTS

The search strategy revealed 2417 articles. Titles and abstracts of 1603 papers were screened against the inclusion and exclusion criteria after removing of duplicates. In all, 134 full-text articles were obtained and screened against inclusion criteria, at which point a further 21 articles were excluded. In the final review, 113 papers were included (Figure 1).

From the 113 papers, the number of participants in the included studies ranged from 14 to 1128 (see Appendix S2). The age of the children in the included studies ranged from birth to 16 years of age. Different types of study designs were retrieved from the research. As can be seen from Appendix S2 and Table S2, most of the included studies were of a cross-sectional study design, which accounted for 32.7% of the total papers. Next came case-controlled studies (14.2%), comparative studies (13.3%), prospective studies (2.7%), observational studies (1.8%), interventional studies (0.9%) and retrospective studies (0.9%). However, 33.6% of papers appeared not to report the type of the study design and this has implications for the quality of the research.

The studies within this systematic review were conducted in 30 different countries; the majority of the research was carried out in Brazil (22.1%) and India (20.4%) (Figure S1). Although most of the research reporting oral health research and children with disabilities were from Brazil, the research studies which were with and actually involved them (category 4b) were from India n = 2, Sweden n = 2, Saudi Arabia n = 1, Turkey n = 1 and Hong Kong n = 1.
A wide range of disabilities were included in this systematic review. For example, intellectual disabilities (which encompass mental disability, developmental disabilities, learning disabilities, mental retardation and cognitive impairment), plus sensory disabilities were included as illustrated in Table S3. The majority of the studies (n = 90, 79.6%) recruited children who had the same type of disability, while (n = 23, 20.4%) studies included children with multiple disabilities.

Of the 113 papers, 10.6% used the word “retarded” to describe children with disabilities, 37.2% “healthy” and 30.9% “normal” to describe nondisabled peers when comparing them with children with disabilities. About 1% used the word “disorder” to describe children with disabilities alongside language which medicalized children in terms of their impairment by placing the condition first and the child last, for example cerebral palsy children, autistic spectrum disorder children, mentally disabled children, Down syndrome children or handicapped children.

3.1 | Research with children

Of the 113 included articles, only 7 (6.2%; Table S1) were categorized as research with children. However, none of them viewed children with disabilities as active participants sharing power and responsibility for the research design and process, in other words they were not involved in the design and conduct of research nor was the child’s perspective taken into consideration. These 7 papers were categorized as the fourth category, which was also classed as research with children, but where children with disabilities were seen as the subjects of research. In these articles, children with disabilities were involved by completing measures, which were wholly designed by adults (category 4b) without children’s input into the wording or format of the measures. These papers used a close-ended oral or written questionnaire answered by children regarding their oral health, dental hygiene habits or the dietary habits.26,27

3.2 | Use of proxies

Of 113 included studies, 47 (41.6%) used proxies to gain the view of children with disabilities. These reports used parents/caregivers, and none of them used clinicians as a proxy. Children in studies including parents/caregivers (category 5a) were either too young (<6 years old) or older with a profound level of intellectual disability or communication was difficult, even with support.

3.3 | Research on children

Finally, as can be seen in Table S1, over half of the included studies, 59 (52.2%) were classified as research on children with disabilities,
where children were not heard or listened to, but only seen (category 6). These articles emphasized that children with disabilities are the objects of research, because they do not attempt enable their participation or include their perspectives or that of their parents.

4 | DISCUSSION

This systematic review presents a hierarchy of the participation of children with disabilities in oral health research ranging from full participation in the research process to no participation. When these classifications were applied to the identified and collected published articles about the oral health of children with disabilities from 2001 to 2017, most studies used children with disabilities as objects with no other participation. This study raises the importance of conducting more studies in the future with children with disabilities to capture their own experiences of oral health and service provision.

This compares with the results of the systematic review of adults with intellectual disabilities which identified that the majority of articles (82%) viewed them as objects in oral health research.\textsuperscript{16,18} It also contrasts with the results of the previous systematic reviews of children in dental research which suggest that children have become increasingly involved in oral health research.\textsuperscript{16,18} These studies found an increase of 17.4% in 2006 to 2014 compared with 7.3% from 2000 to 2005 and a decrease of 87.1% to 64.2% in the number of studies viewing children as objects in oral health research was also found.\textsuperscript{16,18} This indicates that since the first systematic review, oral health research has moved more towards research with children rather than on them. In contrast, our findings suggest the opposite concerning the position of children with disabilities in oral health research. This may be indicative of the way dental research considers children with disabilities as somehow different from their peers because the 2 groups of children appear not to be treated equally in research. It also suggests that disability may be a reason for their exclusion rather than their age. We can further argue that diversity amongst children appears to be neglected, and there is an assumption that they are all the same. If we consider children as a diverse body it makes an important contribution towards the promotion of equality. By failing to include children with disabilities in oral health research, we are denying them a voice. This contradicts the values behind the social model of disability, which sees society as the cause of disability because it erects barriers to participation and this position is supported by the aims of the UNCRC.

In terms of equality and diversity and with the introduction of the Equality Act (2010), it may be claimed that all research with children has a duty to treat children with disabilities equally and protect them from discrimination. Failing to include children with disabilities in consultations, planning and research along with their peers might well be construed as discrimination as case law gradually becomes established. Indeed, assumptions about a child’s competence or ability to take part in research can contribute to both their social and political vulnerability because we are justifying their exclusion from decision-making about their lives.\textsuperscript{28} In terms of health, including oral health, failing to include children and young people in research about them means that we are also failing to recognize their diversity as well as denying them a voice which adds to their existing marginalisation in society.

The most striking finding is that none of the reports involved children with disabilities as active participants sharing power and responsibility for the research process and design. Only 6.2% of the articles were categorized as research with children, but where children with disabilities were seen as the subjects of research. Children in these articles were not involved in the conduct of the research but were asked to complete measures, which were wholly designed by adults (category 4a), such as structured questionnaires regarding children’s oral health, dental hygiene habits or the dietary habits.\textsuperscript{26,27} The perspectives of children with disabilities had not been considered in the research design.

Two of the articles in this systematic review demonstrated that children with disabilities could participate in research through the use of inclusive research methods.\textsuperscript{27,28} In the first study, a questionnaire regarding the oral health practices and knowledge of children with visual impairments was conducted verbally. In the paper by Al-Sarheed et al.\textsuperscript{29} self-perception and need for orthodontic treatment for children with visual (VI) and hearing (HI) impairments were determined by asking each child to rate his/her dental attractiveness. This was performed using the 10-point aesthetic component IOTN for the control group and HI group, while 4 tactile graphics were used for the VI group. Although in these studies, there was a particular focus on children with hearing impairments which might be a common choice made by clinical researchers who possibly understand this as amongst the “easiest” disabled group to work with; they might also be examples of the importance of using inclusive methods in research. The methods were both matched and adapted to the abilities of children; this enabled their voices to be heard and included their views and perceptions.

We can discuss the exclusion of children with disabilities in research in different ways. Perhaps, they are simply ignored as potential contributors, which suggests that they are not viewed as possessing agency.\textsuperscript{30} They might be unseen in some societies because of their absence from community activities and schools, and therefore more difficult to recruit. Furthermore, the adult gatekeeper may suppose that children with disabilities are unable to express their views or have nothing to say. In this situation, children are being judged according to their disability, rather than their ability.\textsuperscript{31} Finally, including them in research might be considered as more time-consuming, expensive, methodologically and ethically challenging, and it also requires particular skill sets on the part of the researcher.

About 41.6% of the reviewed articles involved parents/carers as proxies for children with disabilities and none of them used clinicians (category 5b). This increase in using proxies for children with disabilities suggests that dental researchers may assume that they are unreliable contributors. Although we cannot rely on adult proxy reports of the experiences of children with disabilities alone,\textsuperscript{20} we can acknowledge that parents are skilled interpreters of their children’s signals\textsuperscript{32} and as such researchers may be using the person that knows the child the best to elicit views. Previous research does indicate that the interactional skills of parents and the use of inclusive research methods, influences
successful communication with children with disabilities. We can suggest here that utilizing parents as proxies should complement the views of children with disabilities rather than be used as a substitute. Using proxies might not reflect children’s experiences of oral health and this has already been acknowledged as a limitation in one of the articles included in this systematic review. We can also suggest that parents as proxies might not reflect children with disabilities rather than be used as a substitute. Using language that affords dignity and respect for children might be addressing their own agendas and again we exclude children and lose the impact of their voices.

One of the included articles reported that the oral hygiene data for children with autism were obtained from their parents/caregivers, while the data for children without autism were obtained from the children themselves. Researchers in this article may have assumed that children would be difficult to understand or unable to contribute, and there is a generalized perception in some of the articles that children with autism have “difficulty interacting with people and understanding and following instructions” (p. 44). This discriminates against children with disabilities when compared to their peers in oral health research and is in tension with a rights-based approach to research. The rights of children with disabilities to be consulted and included are reinforced by the social model of disability which promotes participatory research. Other researchers are critical of an approach that reifies the division between children with and without disabilities, suggesting that this division has merely worked to marginalize children with disabilities in research, practice, services and policy.

Most of the included papers, 59 (52.2%) presented research where children with disabilities were not heard or listened to but only seen (category 6). These papers were predominantly aimed at surveying dental health status, estimating treatment needs and comparing the oral health status of children with and without a disability. In these papers, children with disabilities are mainly seen as passive objects rather than as active participants in the research process. An example of research on children with disabilities is observed in one study, which aimed to assess and compare the oral health status, and treatment needs of children with and without disabilities, this was achieved by conducting an oral clinical examination for the children. These 59 papers indicate that children with disabilities were not consulted in any way, illustrating the importance of using inclusive research methods to enable participation in oral health research, simultaneously valuing their views.

The largest volume of research relating to the oral health of children with disabilities originated from Brazil followed by India. This might simply reflect a high number of active researchers and dental institutes within these countries. In contrast, there might be other facilitators encouraging the conduct of research studies such as lack of bureaucratic, ethical and governance processes and funding availability. Interestingly, there were also differences in the countries where the study involved children with disabilities in the research process. These (category 4b) were from India n = 2, Sweden n = 2, Saudi Arabia n = 1, Turkey n = 1 and Hong Kong n = 1. The single most striking observation to emerge from comparing this finding with the previous systematic reviews was that none of the papers originated from the UK where children were actively involved throughout the research process. The reason might be because researchers in the UK have involved children with disabilities as a part of the category of child and not considered the diversity of the group, although we would expect to see a range of inclusive methods in the studies to ensure participation by all children. We can suggest here that there appears to be enormous scope for international collaborative research within the field of oral health with children with disabilities.

The type of language used in many of the included articles to describe children with and without disabilities is unacceptable with comparisons between “retarded” and “normal” or “healthy” children. Referring to children without disabilities as “normal children” or “healthy children” implies that children with disabilities are abnormal, deviant, strange or unhealthy which reflects the medical model of disability, individualizing disability and placing it within the person. Terms that medicalize children such as “the Down’s syndrome child” or the “cerebral palsy child” are also unacceptable because they objectify and dehumanize children, undermining their worth. These terms and ways of viewing children with disabilities are also considered negative and offensive by people with and without disabilities alike. Using language that affords dignity and respect for children with disabilities, viewing them as active individuals with agency should be advocated in future oral health research.

4.1 Limitations of the study

It is acknowledged that the current systematic review has some limitations. The search was limited to the English language, 3 main electronic databases and dental journals for practical reasons and because they mirror the designs of the previous systematic reviews. This means that some related studies may have been overlooked, particularly those written in languages other than English. Another limitation is the problematic use of the term “case-control” in the studies which is poorly defined. For example, studies where the oral health of children with disabilities is compared to those without should not strictly speaking be defined as “case-control.” This is because cases of disease are not identified and matched with controls that do not have the disease and then supposed risk factors for the disease explored. Instead, they are poorly designed cross-sectional studies. These study classifications are those of the original authors and not our own.

5 CONCLUSION AND RECOMMENDATIONS

The results of this review suggest that in the majority of oral health research, children with disabilities are seen, but not heard or listened to. Future research should attempt to involve children with disabilities in oral health research by designing studies using innovative methodologies, drawn from different disciplines, as far as possible. This aims towards a rights-based approach in research to avoid the discrimination and disempowerment of children with disabilities by including their voices in research that concerns them. In this way, we can ensure that their perspectives are obtained through not only listening to but also acting upon their voices to improve their oral health outcomes.
REFERENCES