Oral health status of children and adolescents with intellectual disabilities: a systematic review and meta-analysis

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PUBLICATION DATA
Accepted for publication 19th April 2017.
Published online 19th June 2017.

ABBRVIATIONS
DFS Decayed and filled surfaces on permanent teeth
DFT, dt Decayed and filled permanent, primary teeth
DMFS Decayed, missing, and filled surfaces on permanent teeth
DMFT, dft Decayed, missing, and filled permanent, primary teeth
PCO Population, intervention/interest, comparator, outcome
SMD Standardized mean difference

AIM To compare the oral health status of children and adolescents affected by intellectual disabilities with their unaffected counterparts.

METHOD Citations published in English were searched from electronic databases (PubMed, Embase, Web of Science, and Scopus) from their start dates to March 2017. The whole process was conducted following PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analyses) guidelines. The PICO (population, intervention/interest, comparator, outcome) principle was used to formulate the topic. Studies were synthesized through qualitative summary or, whenever possible, meta-analysis.

RESULTS The initial search yielded 2393 records. Thirty-nine studies from 22 countries were identified for qualitative analysis; 26 studies were eligible for meta-analysis. Participants with intellectual disabilities had higher levels of dental plaque, worse gingival status, fewer decayed and filled permanent teeth, and similar caries experience between males and females. These findings were supported by both qualitative and quantitative analysis. Various patterns of caries experiences were indicated by qualitative analysis, but it was not substantiated by meta-analysis.

INTERPRETATION There is increasing worldwide interest in oral health status of children with intellectual disabilities. Differences in dental plaque deposition, gingival inflammation, and the number of decayed and filled permanent teeth were investigated between children and adolescents with and without intellectual disabilities. Evidence remains elusive about the pattern of caries experience among those children.

Individuals diagnosed with intellectual disabilities have limitations in both intellectual functioning and adaptive behaviour. The American Association on Intellectual and Developmental Disabilities defines adaptive behaviour as ‘the collection of conceptual, social, and practical skills that all people learn to function in their daily lives’.[1] This broad concept covers a wide range of aspects such as personal care, health care, and application of basic skills (conceptual, social, and practical skills) in daily life.

Tooth brushing, a universally accepted personal care practice, is also one aspect of adaptive behaviour. Children with intellectual disabilities have been reported to have more obstacles in daily tooth brushing than healthy children. This might be due to their difficulty in achieving the required manual dexterity for self-care and lack of awareness of the importance of oral hygiene practice.[2] Poor oral health status among them has been noted. Mounting evidence suggests that, compared with the typically developing population, individuals with intellectual disabilities had poorer oral hygiene status,[1–5] worse periodontal condition,[3,4,6,7] and higher risk of developing caries.[5,8–10] Kavvadia et al. observed higher levels of dental plaque among adolescents and young adults with intellectual disabilities, and more severe intellectual impairment was associated with worse periodontal status.[11] Oliveira et al. demonstrated that the individuals with mental impairment had not only poorer oral health status but also more difficulty in getting access to oral health services than their siblings without intellectual disabilities.[12] Children with intellectual disabilities have reduced ability to understand new information and complex instructions.[13] They have been reported to be less cooperative with dentists and have more problems with the management of dental behaviour.[14] Consequently, oral health care and routine dental treatment become much more challenging for them, resulting in poorer oral health and rapid progression of existing oral diseases.[15]

Limited access to dental care and compromised ability in self-care make oral health promotion particularly important for patients with intellectual disabilities. However, conflicting evidence exists about the oral health status of children and adolescents with intellectual disabilities. Although several studies have found poorer oral health status among these populations,[11,12] other studies have
observed that children with intellectual disabilities exhibited fewer caries, less dental plaque deposition, and better gingival condition than children without intellectual disabilities. The issue is further complicated by claims that caries experience and oral hygiene status among children with intellectual disabilities are similar to children without intellectual disabilities.

Understanding oral health status among children and adolescents with intellectual disabilities is essential for effectively promoting their oral health. This systematic review aimed to summarize existing evidence in order to compare oral health status (dental plaque deposition, periodontal status, and caries experience) between children and adolescents with and without intellectual disabilities.

METHOD

Topic and search strategy
The focused question governing the systematic search was formulated by PICO principles, which refers to four elements: population, intervention/interest, comparator, and outcome. Thus, the topic was, ‘Do children and adolescents (P) affected by intellectual disabilities (I), compared with their unaffected counterparts (C), have worse oral health status (O)?’

Four electronic databases, PubMed, Embase, Web of Science, and Scopus, were searched from their start date to March 2017, using automatic e-mail alerts. To reduce the publication bias, ‘grey literature’ – work ongoing but not published – was checked on the Clinical Trials website (http://clinicaltrials.gov), which accommodates registered observational studies. The number of those studies has increased in recent years. The search strategy consisted of terms and key words relating to oral health (‘dental plaque’, ‘dental caries’, ‘gingival status’) and ‘intellectual disabilities’ as used in previous systematic reviews on oral hygiene and intellectual disabilities. The target population of ‘children and adolescents’ was limited to ‘age below 18 years’ (Appendix S1, online supporting information).

Study selection and eligibility criteria
This systematic review was performed in accordance with the PRISMA (Preferred Reporting Items for Systematic reviews and Meta- Analyses) statement (Fig. S1, online supporting information). Initially, titles and abstracts of searched records were screened by two authors independently to identify ‘potentially eligible’ studies. Full texts of ‘potentially eligible’ studies were retrieved to determine whether all inclusion criteria were met. The identified ‘eligible’ studies formed the basis of the qualitative analysis.

Quality assessment
The quality of the studies that formed the basis of meta-analysis was assessed by the Newcastle–Ottawa Scale (NOS). A ‘star system’ was developed to evaluate studies from three domains: selection (four items), comparability, and outcome/exposure (three items). A maximum of one star for each item was awarded to ‘selection’ and ‘outcome/exposure’. A maximum of two stars was given to ‘comparability’.

Data extraction and analysis
Information extracted from eligible studies included year of publication, participants, sample size, age range, key outcome measurements (plaque indices, gingival indices, and caries indices), and main findings (comparison between groups and p-values). Continuous data were reported as mean and standard deviation or median and interquartile range. Categorical data were described by proportion and frequency. When p-values or proportion values were not reported in the original studies, they were indirectly derived through GraphPad QuickCalcs (GraphPad Software, Inc., La Jolla, CA, USA) whenever possible. Meta-analysis and assessment of publication bias were performed by Stata (StataCorp. Stata Statistical Software. Release 12. College Station, TX: StataCorp LP). Subgroup analyses were performed when various indices were used to indicate the same outcome measure being combined. When results from three or more studies were to be pooled, a random-effects model was employed since each study included in this meta-analysis provided information about a different effect size.

RESULTS

Study selection and characteristics
The initial search from online electronic databases yielded 2393 records, and 1629 remained after removal of duplications. The screening of titles and abstracts resulted in 76 potentially eligible studies, among which 39 eligible studies (reported from 41 articles) were identified and formed the basis of qualitative analysis (24 studies assessed dental plaque, 21 studies assessed gingival or
periodontal status, and 32 studies assessed caries status. Twenty-six studies were suitable for meta-analysis (12 studies on dental plaque, 8 on periodontal status, and 23 on caries). The kappa value of interexaminer agreement was 0.897 (Fig. S1).

The included studies were published within the past 50 years. Three studies were published before 1985 and 32 were published after 2000. There were 19 studies published within 5 years. These studies came from 22 countries: five studies each from Brazil and India, three each from Saudi Arabia and Japan, and one or two studies from each of several other countries (Table SI, online supporting information).

The included studies recruited a total of 56,417 participants, with 33,25 participants in the case group, and 53,092 in the comparison group. Four studies recruited children under 6 years (primary dentition), 32 studies recruited children and adolescents aged 1 to 18 years (primary dentition, mixed dentition, and permanent dentition), and three studies recruited adolescents aged 14 to 18 years (permanent dentition) (Table SI).

**Quality assessment**

Quality assessment was performed according to the ‘star system’ of NOS. The total number of stars ranged from 1 to 9 for each study. Among the 26 studies included for meta-analysis, 20 (77%) were rated as having at least 7 stars. The mean number of stars was 3.2 (maximum of 4) for the ‘selection’ category, 1.6 (maximum of 2) for the ‘comparability’ category, and 2.3 (maximum of 3) for the ‘outcome/exposure’ category (Table SII, online supporting information).

**Dental plaque**

Twenty-four studies assessed the severity of dental plaque accumulation using the following indices: the Simplified Oral Hygiene Index (16 studies), Plaque Index of Silness and Löe (five studies), Patient Hygiene Performance Index of Podshadley and Haley (one study), Visible Plaque Index of Ainamo and Bay (one study), oral hygiene partial scoring modification described by Ramford (one study), and an unspecified oral hygiene index (one study) (Table SIII, online supporting information).

Qualitative analysis of dental plaque was based on 24 studies between children with and without intellectual disabilities. Three studies reported that children with intellectual disabilities had lower oral hygiene index scores. More than half of the studies supported the idea that children and adolescents affected by intellectual disabilities had significantly more dental plaque than their unaffected counterparts.

Twelve studies with 14 comparisons were eligible for quantitative analysis. Subgroup analysis was performed, because different plaque indices, the Simplified Oral Hygiene Index of Greene and Vermillion and the Plaque Index of Silness and Löe, were used by the eligible studies. There was no statistical difference in dental plaque accumulation between children with and without intellectual disabilities as measured by the Simplified Oral Hygiene Index of Greene & Vermillion (z=1.69; p=0.091; standardized mean difference [SMD]=0.60, 95% CI 0.10–1.30). In contrast, the Plaque Index of Silness and Löe showed that children and adolescents with intellectual disabilities had higher mean plaque scores than those without intellectual disabilities (z=4.38; p=0.000; SMD=0.88, 95% CI 0.49–1.27). The overall effect demonstrated that children with intellectual disabilities had significantly higher levels of dental plaque than those without intellectual disabilities (z=2.62; p=0.009; SMD=0.67, 95% CI 0.17–1.18) (Fig. 1).

**Periodontal status**

Periodontal status among children and adolescents with and without intellectual disabilities was assessed by 21 studies using various indices. The Community Periodontal Index of Treatment Needs, Community Periodontal Index (CPITN/CPI, four studies), probing depth (three studies), and the Modified Gingival Index (two studies) owing to the relatively larger number of studies using these indices. Studies using other indices (the Gingival Bleeding Index of Ainamo and Bay, the Gingival Hyperplasia Index, the modified Papillary-Marginal-Attached Gingiva Index of Massler, the Gingival Index of Löe 1967, the ‘Periodontal Index’, bleeding on probing, and the Visible Periodontal Index) had less impact on the outcome of meta-analysis (Table SIV, online supporting information).

Among 21 studies, one reported that preschool children with intellectual disabilities had lower gingival index scores. One study illustrated that there was no statistical difference in the prevalence of gingivitis among both groups, while children in the case group (78.0%) had significantly more generalized gingivitis than comparison group (20.0%). Most studies supported the observation that children and adolescents with intellectual disabilities demonstrated worse periodontal status.

Meta-analysis was performed using data pooled from nine comparisons in eight studies. There was no statistical difference in periodontal status between children with and without intellectual disabilities as measured by the Gingival Index of Löe and Silness (z=1.88; p=0.06; SMD=0.53, 95% CI –0.02 to 1.08), while the Modified Gingival Index showed that participants with intellectual disabilities had worse periodontal status (z=2.73; p=0.006; SMD=0.97, 95% CI 0.27–1.67). The overall effect demonstrated that children affected by intellectual disabilities had significantly worse gingival status than those without intellectual disabilities (z=2.53; p=0.011; SMD=0.63, 95% CI 0.14–1.12) (Fig. 2).
Dental caries

Outcome measurement

The difference in dental caries experience between children with and without intellectual disabilities was evaluated based on 32 studies. Diagnoses of caries in these studies were mainly performed in compliance with World Health Organization criteria. A total of 26 indices were used to indicate caries experience. For permanent teeth, decayed, missing, and filled teeth (DMFT) was used in 16 studies; decayed, missing, and filled surfaces (DMFS) was adopted in seven studies; decayed and filled teeth (DFT) was reported by two studies; and decayed and filled surfaces (DFS) was reported in one study. For primary teeth, decayed, missing or extracted, and filled teeth (dm(e)ft) was applied to nine studies; decayed, missing, and filled surfaces (dmfs) was reported in five studies; decayed and filled teeth (dft) was reported in six studies; and decayed and filled surfaces (dfs) was reported in one study. For primary teeth, decayed, missing or extracted, and filled teeth (dm(e)ft) was applied to nine studies; decayed, missing, and filled surfaces (dmfs) was reported in five studies; decayed and filled teeth (dft) was reported in six studies; and decayed and filled surfaces (dfs) was reported in one study. Caries experience in primary and permanent teeth were assessed by using DMFT/dmft (four studies), DMFT/dft (one study), DFT/dft (one study), caries-free rate (eight studies), and caries prevalence (five studies). Decayed teeth and decayed surfaces, missing teeth and missing surfaces, and filled teeth and filled surfaces were also investigated separately (Table SV, online supporting information).

Qualitative analysis

DMFT (16 studies), dm(e)ft (nine studies), caries-free rate (eight studies), and DMFS (seven studies) were the most frequently used indices among the included studies. As for DMFT and dm(e)ft, more than half of the included studies supported the idea that there was no significant difference between the case and comparison groups. Nearly half of the relevant comparisons revealed that children with intellectual disabilities had fewer filled permanent teeth, more missing permanent teeth due to caries, higher DMFS score, and lower DMFS score (Table SVI, online supporting information).

Quantitative analysis

A total of 23 studies that reported outcome in means and standard deviations formed the basis of the meta-analysis. Key findings about participants’ caries experience are summarized in the last column of Table SVI. No publication bias existed according to Egger’s test (p>0.05). The meta-analysis suggested that children and adolescents with intellectual disabilities had
fewer decayed and filled permanent teeth (DFT, \( z = 2.80; p = 0.005; \) SMD = -0.36, 95% CI -0.62 to -0.11), while no significant difference was detected in other indices and the overall effect (Fig. S2, online supporting information).

**Distribution of caries by sex**

Caries experiences (DMFT, DMFS, dm(e)ft, dft/DFT) in males and females were reported in five studies. Comparisons of caries experience were performed between males with and without intellectual disabilities (males [case vs comparison]), females with and without intellectual disabilities (females [case vs comparison]), males with intellectual disabilities and females with intellectual disabilities (case [male vs female]), and males without intellectual disabilities and females without intellectual disabilities (comparison [male vs female]). One study found out that both males and females in the case group were vulnerable to dental caries in permanent dentition: females with intellectual disabilities (mean 9.31, standard deviation [SD] 10.40) had significantly higher DMFS scores than females without intellectual disabilities (mean 2.10, SD 4.63, \( p < 0.05 \)); males with intellectual disabilities had higher scores (mean 17.58, SD 24.12) than the comparison group (mean 3.25, SD 3.75, \( p < 0.01 \)).

One study suggested that males in the intellectual disabilities group had lower DMFT (mean 2.82, SD 3.28) than males in the comparison group (mean 4.07, SD 3.51, \( p = 0.034 \)). No significant difference in the caries pattern was reported by other studies, and meta-analysis did not find statistical differences in DMFT or dmft scores among males or females with or without intellectual disabilities (Table SVII, online supporting information).

**Distribution of caries by tooth surfaces**

Caries was described by tooth surfaces in three studies. One study stated that the prevalence of pit and fissure caries was similar between children with (55.2%) and without (52.6%) intellectual disabilities, while the prevalence of smooth surface caries among children with intellectual disabilities (28.5%) was lower (\( p < 0.001 \)) than among children without intellectual disabilities (40.8%). Two studies indicated that children with or without intellectual disabilities shared similar profiles in proximal caries, but fewer decayed proximal surfaces were filled among children and adolescents with intellectual disabilities (Table SVIII, online supporting information).

**DISCUSSION**

**Focused question and study selection**

The World Health Organization handbook for guideline development states, 'PICO refers to four elements that should be in a question governing a systematic search of the evidence: population, intervention, comparator and outcome.'
outcomes. Although the PICO principle was primarily designed for clinical trials and interventional studies, it can also formulate a clear and structured topic when applied to observational studies. In this study, governed by PICO principles, a structured research question was formulated as follows: 'Do children and adolescents (population) affected by intellectual disabilities (interest), compared with their unaffected counterparts (comparator), have worse oral health status (outcome)?'

Citations relevant to this topic were retrieved from four online electronic databases. The eligible studies were conducted in 22 countries with a temporal span of 50 years. All the included articles were observational studies with a case group consisting of individuals with intellectual disabilities and a comparison group comprising participants without intellectual disabilities.

A total of 56,417 participants (<18y) informed this review, with 3325 in the case group and 53,092 in the comparison group. Quality assessment was performed using a 'star system' developed for NOS, and 77 per cent of the eligible studies were awarded at least seven stars. As for the outcome measurements, indices for oral hygiene status, gingival or periodontal status, and caries experience were used for evaluation. To ensure reliability of the outcome measurements, self-reported outcomes were excluded and only primary data collected by the researchers were used for analysis. A total of 1629 records remained after removal of duplications. There were 39 studies from 41 articles identified for qualitative analysis. Among these, 26 were feasible for conducting meta-analysis. The kappa value for interexaminer agreement was 0.897, which indicated almost perfect agreement.

**Overview of oral hygiene status**

Various measurements were used to assess oral hygiene status in 24 studies (Simplified Oral Hygiene Index of Greene and Vermillion, Plaque Index of Silness and Löe, Patient Hygiene Performance Index of Podshadley and Haley, Visible Plaque Index of Ainamo and Bay, oral hygiene partial scoring modification described by Ramfjord, and an unspecific oral hygiene index). Both qualitative and quantitative analysis provided evidence that participants with intellectual disabilities had significantly higher levels of dental plaque than those without intellectual disabilities.

Periodontal status was investigated in 21 studies using 11 indices (Gingival Index of Löe and Silness, CPITN/CPI, probing depth, Gingival Bleeding Index of Ainamo and Bay, Gingival Hyperplasia Index, Modified Gingival Index of Lobene, modified Papillary-Marginal-Attached Gingiva Index of Massler, 'Periodontal Index', bleeding on probe, and Gingival Index of Löe 1967, Visible Periodontal Index). Both qualitative and quantitative analysis suggested that children and adolescents with intellectual disabilities had worse periodontal status than their unaffected counterparts.

Dental caries was evaluated in 32 studies. Diagnostic criteria of caries in eligible studies mainly followed the World Health Organization guideline. A total of 26 indices (DMFT, dm[ef]t, DMFT/dmft, DMFS, dmfs, DFT, dft, DMFT/dft, DFT/dft, DFS, dfs, decayed teeth [primary, permanent, both], decayed surfaces [primary, permanent], missing teeth [permanent], missing or extracted teeth [primary], missing surfaces [primary, permanent], filled teeth [primary, permanent], filled surfaces [primary, permanent], caries prevalence, caries-free rate) were used. Quantitative analysis revealed that children and adolescents with intellectual disabilities had fewer decayed and filled permanent teeth, while qualitative studies indicated that children with intellectual disabilities might have higher DMFS scores, and more missing permanent teeth due to caries, and fewer filled permanent teeth than children without intellectual disabilities. Other researchers demonstrated that individuals with intellectual disabilities had more decayed teeth, fewer filled teeth, and higher numbers of missing teeth due to caries, which was partly due to their difficulty in getting access to oral health care services. Evidence revealed that the number of filled proximal surfaces was lower among children and adolescents with intellectual disabilities. This indicated the limited dental care facilities for children and adolescents with special needs. Restoration of proximal surfaces is more difficult than that of occlusal or smooth surfaces. Uncooperativeness of individuals with intellectual disabilities presents a further challenge for normal dental treatment procedures.

The existing evidence on oral health status of children and adolescents with intellectual disabilities is inconclusive. This might be associated with the large temporal span in years of publication of the eligible studies. A study published in 1967 claimed that children with intellectual disabilities had more caries and worse periodontal status, which indicated their difficulty in getting access to dental services at that time. After decades when few relevant studies could be identified, increasing numbers of studies emerged from the 2000s onwards. Recent studies have suggested that individuals with intellectual disabilities have a similar oral hygiene status to those without intellectual disabilities. A few studies have even claimed better oral health status among individuals with intellectual disabilities. Improvement in the oral health status of patients with intellectual disabilities may be associated with increasing awareness of the importance of oral health and the increasing number of oral health promotion activities.

**Limitations**

One limitation of this meta-analysis is that the total number of children without intellectual disabilities (n=53,092) was 16 times that of children with intellectual disabilities (n=3325). This might reduce the power to detect the true effect size between the two groups. However, this limitation was inherited directly from the original studies, which had formed the basis of this study. The sample sizes in the comparison group were not matched with the case group in half of the included studies, while the remaining studies recruited relatively small numbers of participants. The limited number of participants in the case group may have
been due to the difficulty of patients with intellectual disabilities in cooperating with examiners. Meanwhile, it has been reported that there was a significant positive association between the severity of oral disease and that of intellectual disabilities. However, patients with severe intellectual disabilities may not tolerate dental examinations, and findings of eligible studies might therefore reflect the oral health status of patients with less severe intellectual disabilities. Clinical data from patients with various levels of intellectual disabilities are needed to provide a more thorough understanding of the oral health status of children and adolescents with intellectual disabilities.

Meta-analysis was developed to summarize the results of different research studies in relevant fields. However, it has its own limitations or critics, especially for observational studies. The summary estimates synthesized by a meta-analysis do not mean that the meta-analyses are able to give a convincing answer to the particular clinical question. Substantial collaborations with researchers, clinical practitioners, and statistical experts are still needed to address the specific question.

**Conclusion**

There has been growing worldwide interest in the oral health status of individuals with intellectual disabilities since the 2000s. Children and adolescents with intellectual disabilities had higher level of dental plaque, worse gingival status, and fewer decayed and filled permanent teeth than those without intellectual disabilities. Caries experience between males and females was similar in participants with or without intellectual disabilities. Although qualitative analysis suggested that children with intellectual disabilities had higher DMFS scores, more missing permanent teeth due to caries, and fewer filled permanent teeth than children without intellectual disabilities, these were not substantiated by quantitative analysis. Existing evidence on the profiles of specific caries experience among children and adolescents with intellectual disabilities remains elusive. More well-designed studies on children and adolescents with different levels of intellectual disabilities are warranted.

**ACKNOWLEDGEMENTS**

The authors gratefully acknowledge Samantha Kar Yan Li (Senior Technical Officer, Centralized Research Laboratories, Faculty of Dentistry, The University of Hong Kong) for her advice on statistical analysis. The authors have stated that they had no interest that could be perceived as posing a conflict or bias.

**SUPPORTING INFORMATION**

The following additional material may be found online:

- **Figure S1**: PRISMA flow diagram of study selection.
- **Figure S2**: Meta-analysis of caries experiences between children with and without intellectual disabilities.
- **Table S1**: Characteristics of eligible studies on oral health status of children and adolescents with and without intellectual disabilities.
- **Table SII**: Quality of 26 studies assessed by Newcastle–Ottawa Scale.
- **Table SIII**: Dental plaque status among children/adolescents with and without intellectual disabilities.
- **Table SIV**: Gingival/periodontal status among children/adolescents with and without intellectual disabilities.
- **Table SV**: Caries experience among children/adolescents with and without intellectual disabilities.
- **Table SVI**: Caries experience of males and females among children and adolescents with and without intellectual disabilities.
- **Table SVII**: Caries experiences in various tooth surfaces.
- **Appendix S1**: Search strategy.

**REFERENCES**


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