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REVIEW ARTICLE



# Does dental agenesis have an impact on OHRQoL of children, adolescents and young adults? A systematic review

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**ABSTRACT**

Dental agenesis (DA) is defined as the congenital absence of teeth and is considered as the most common dental anomaly. It may cause speech and masticatory dysfunctions as well as esthetic problems. Its impact on oral health-related quality of life (OHRQoL) is not fully understood. The aim of the study was to assess whether DA affects OHRQoL of children, adolescents and young adults. A broad search was done on databases (Pubmed, Scopus, Web of Science and Virtual Health Library) using Medical Subject Headings (MeSH) and free terms. Eligibility criteria for article selection were predetermined and were classified according to quality assessment and risk of bias. The electronic search produced 178 titles and abstracts. After excluding duplicate abstracts and applying the eligibility criteria, three articles were assessed for the final qualitative synthesis. The three articles were classified as moderate quality and present risk of bias. No articles were found that had evaluated children and young adults. From the three articles that were selected, only one was found to have a greater impact in the adolescent agenesis group with statistical differences in all domains. There is insufficient evidence available to conclude if DA affects OHRQoL of children, adolescents and young adults.

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Dental agenesis; quality of life; oral health; child; adolescent; young adult

## Introduction

Dental agenesis (DA) is defined by the absence of development of one or more teeth [1], and is considered to be the most common dental anomaly [2]. It can affect both deciduous and permanent dentitions [2]. In the deciduous dentition, it has a prevalence varying between 0.2% [3] and 2.38% [4], while in the permanent dentition, it has a prevalence between 2.4% [5] and 13.3% [6], when third molars are excluded. Alterations in number of teeth result from disorders during the initiation stage and proliferation of dental development. These disorders may be due to failure in the process of dental laminin induction and tooth budding (initiation period) or deficiency in cell multiplication, which promotes the development of tooth buds (proliferative period) [7].

Although dental agenesis is not highly prevalent [6], it may cause speech and masticatory dysfunctions as well as esthetic problems [8], which can affect the individual's social life. A smile plays an important role in the lives of adults, adolescents and children, and is important for their overall emotional well-being. In addition, having a perfect smile is related to physical attraction, which plays an important role in how we see ourselves, how we feel, and how we are seen by others [9]. The dental aspect of health promotion and the relationship between oral health and quality of life has been the focus of dentistry professionals [10], mainly due to the relevance of oral problems and the physical and psychosocial impacts.

The impact of DA on oral health-related quality of life (OHRQoL) has received little attention in the literature. No previous systematic review has investigated the impact of DA on OHRQoL in children, adolescents and young adults. This dental anomaly and its impact on well-being and quality of life should be assessed [11]. This systematic review aims to test the hypothesis that DA impacts on the OHRQoL of children, adolescents and young adults.

## Materials and methods

### Protocol and registry

The protocol of this systematic review was based on the PROSPERO database (PROSPERO registry number: CRD 42017060451). It was conducted following the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) [12].

### Focused question

Does dental agenesis impact on the OHRQoL in children, adolescents and young adults?

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### Search strategy and studies selection

A broad search was conducted up to 2 February 2017 using the following electronic bibliography databases: PubMed, Scopus, Web of Science and Virtual Health Library (VHS). The VHS presented articles in English and other languages (Lilacs, Scielo). There were no restrictions on language, or filters or publication dates. The search strategy included descriptors selected from a combination of a previous search in Medical Subject Headings (MeSH) and the free terms as described in Table 1. A complementary manual search was also performed by screening the references of the selected articles to find any that did not appear in the database search. The grey literature was also consulted through Opensigle (http://www.opengrey.eu) and researchers were contacted to identify unpublished and ongoing studies.

Initially, three of the authors independently (ASR, JSF and LAA) selected the studies by titles and abstracts where all articles are screened for relevance. Articles appearing in more than one database were considered only once. The relevant abstracts were then retrieved and read. After that the full articles or manuscripts of the selected papers were retrieved, and the final selections for inclusion were made. When the titles and abstracts were not clear, the articles were accessed in their entirety. Disagreement between the three authors was resolved by consensus or by a fourth reviewer (LSA).

**Table 1.** Search strategy.

| Database | Search strategy |
|---|---|
| *PubMed* | **#1** (((((((Anodontia[MeSH Terms]) OR Anodontia[Title/Abstract]) OR Tooth Ageneses[Title/Abstract]) OR Tooth Agenesis[Title/Abstract]) OR Dental agenesis[Title/Abstract]) OR Hypodontia[Title/Abstract]) OR Oligodontia[Title/Abstract]) OR Tooth Abnormalities[Title/Abstract] Filters: Publication date from 2017/02/15<br>**#2** ((((((((((Quality of Life[MeSH Terms]) OR Quality of Life[Title/Abstract]) OR Oral health impact profile[Title/Abstract]) OR Oral health related quality of life[Title/Abstract]) OR OHIP[Title/Abstract]) OR QoL[Title/Abstract]) OR OHRQoL[Title/Abstract]) OR ECOHIS[Title/Abstract]) OR Child-OIDP[Title/Abstract]) OR COHIP[Title/Abstract]) OR CPQ[Title/Abstract]) OR OIDP[Title/Abstract]) OR SOHO-5[Title/Abstract] Filters: Publication date from 2017/02/15<br>**#3** (((((((Child[MeSH Terms]) OR Adolescent[MeSH Terms]) OR Young adult[MeSH Terms]) OR Child[Title/Abstract]) OR Children[Title/Abstract]) OR Adolescents[Title/Abstract]) OR Young adults[Title/Abstract] Filters: Publication date from 2017/02/15<br>**#1 AND #2 AND #3** |
| *Scopus* | **#1** ( TITLE-ABS-KEY ( anodontia ) OR TITLE-ABS-KEY ( tooth AND ageneses ) OR TITLE-ABS-KEY ( tooth AND agenesis ) OR TITLE-ABS-KEY ( dental AND agenesis ) OR TITLE-ABS-KEY ( hypodontia ) OR TITLE-ABS-KEY ( oligodontia ) OR TITLE-ABS-KEY ( tooth AND abnormalities )<br>**#2** ( TITLE-ABS-KEY ( quality AND of AND life ) OR TITLE-ABS-KEY ( oral AND health AND impact AND profile ) OR TITLE-ABS-KEY ( oral AND health AND related AND quality AND of AND life ) OR TITLE-ABS-KEY ( ohip ) OR TITLE-ABS-KEY ( qol ) OR TITLE-ABS-KEY ( ohrqol ) OR TITLE-ABS-KEY ( ecohis ) OR TITLE-ABS-KEY ( child-oidp ) OR TITLE-ABS-KEY ( cohip ) OR TITLE-ABS-KEY ( cpq ) OR TITLE-ABS-KEY ( oidp ) OR TITLE-ABS-KEY ( soho-5 ) )<br>**#3** ( TITLE-ABS-KEY ( child ) OR TITLE-ABS-KEY ( children ) OR TITLE-ABS-KEY ( adolescents ) OR TITLE-ABS-KEY ( young AND adults ) )<br>**#1 AND #2 AND #3** |
| *WOS* | **#1** ((((((TOPIC:(anodontia) OR TOPIC: (Toothagenesis)) OR TOPIC: (Tooth Agenesis)) OR TOPIC: (Dental agenesis)) OR TOPIC:(Hypodontia)) OR TOPIC:(Oligodontia)) OR TOPIC: (Tooth Abnormalities))<br>**#2** ((((((((((TOPIC:(Quality of Life) OR TOPIC: (Oral health impact profile)) OR TOPIC:(Oral health related quality of life))OR TOPIC: (ohio)) OR TOPIC:(QoL)) OR TOPIC: (optqol)) ORTOPIC: (echis)) OR TOPIC: (Child-odp)) OR TOPIC: (conip)) ORTOPIC: (CPQ)) OR TOPIC: (odp))OR TOPIC: (SOHO-5))<br>**#3** TOPIC: (Child) OR TOPIC: (Children) OR TOPIC: (Adolescents) OR TOPIC: (Young adults)<br>**#1 AND #2 AND #3** |
| *VHL* | (tw:(Anodontia)) OR (tw:(Tooth Ageneses)) OR (tw:(Tooth Agenesis)) OR (tw:(Dental agenesis)) OR (tw:(Hypodontia)) OR (tw:(Oligodontia)) OR (tw:(Tooth Abnormalities)) AND (tw:(Quality of Life)) OR (tw:(Oral health impact profile)) OR (tw:(Oral health related quality of life)) AND (tw:(Child)) OR (tw:(Children)) OR (tw:(Adolescents)) OR (tw:(Young adults)) |

### Selection criteria

The inclusion criteria were based on the population, exposition, comparisons and outcomes (**PECOS**) format, as follows:

Population (**P**): children, adolescents and young adults, according to WHO [13] (children: 2 to 10 years; adolescents: 10 to 19 years; and young adults: 19 to 24 years).

Exposition (**E**): dental agenesis.

Comparison (**C**): the case group with dental agenesis should be compared to control groups without agenesis.

Outcome (**O**): oral health detected by validated quality of life instruments.

Study design (**S**): cross-sectional studies with control group, case-control studies.

The exclusion criteria were studies with participants out of the age range or with medical conditions (e.g. systemic disorders, syndromes and congenital malformations). In addition, studies that evaluated the impact of dental agenesis with other dental anomaly (without separate results) or assessed only the psychometric properties of OHRQoL instrument were excluded. Furthermore, papers out of the proposed theme, editorial letters, pilot studies, literature reviews, theses, observational and descriptive studies, such as case reports and case series were also excluded.

### Quality assessment and control of bias

After the inclusion of the abstracts that fulfilled the selection criteria and verification of their eligibility by reading the complete articles, the studies were submitted for the quality assessment. The methodological quality assessment and control of bias of the studies were independently evaluated by two authors (LAA and ASR). The identified manuscripts were evaluated, and if there were differences between the two readers, they were resolved through consensus with all authors (ASR, JSF, LSA, LAA). If relevant data were missing, the authors of the articles in question were contacted for additional information. Quality assessment and bias risks were carried out according to the guidelines described by Fowkes and Fulton [14].

The Fowkes and Fulton [14] quality assessment allows classifying of cross-sectional studies, cohort, controlled-clinical trial and case-control studies. This guideline presents

questions about study design, sample representativeness, control group characteristics, quality measurements and results, and distortions. By checking each item from the guide, the importance of failure effects on the results was scored as major problems (++) or minor problems (+), and a decision regarding the accuracy of producing useful information was made. Items where the question was not applicable were marked 'NA'.

Therefore, studies without problems, studies only checked with minor problems and those checked with major problems associated or not with minor problems were identified as of high, moderate and low scientific evidence, respectively. Confounding factors and risk of bias were also identified by three specific questions presented at the end of the guideline using the option 'yes' or 'no' for answer. If a problem ('yes') was identified in one of these three questions, it was considered a study with a risk of bias.

### *Data collection*

The data from the included papers were compiled, and the following data extracted: author(s), year, country, study design, groups evaluated, sample size, sample age, age range, gender, instrument applied and outcomes.

## Results

Figure 1 shows the flowchart (PRISMA) that describes the number of articles identified at each step of the study. The search strategy initially identified 178 articles in the electronic databases of which 67 were excluded, because they were duplicates. We did not identify unpublished and ongoing studies in the grey literature. After the analysis of the titles and abstracts following the eligibility criteria, 105 articles were excluded. After analysing the complete texts of

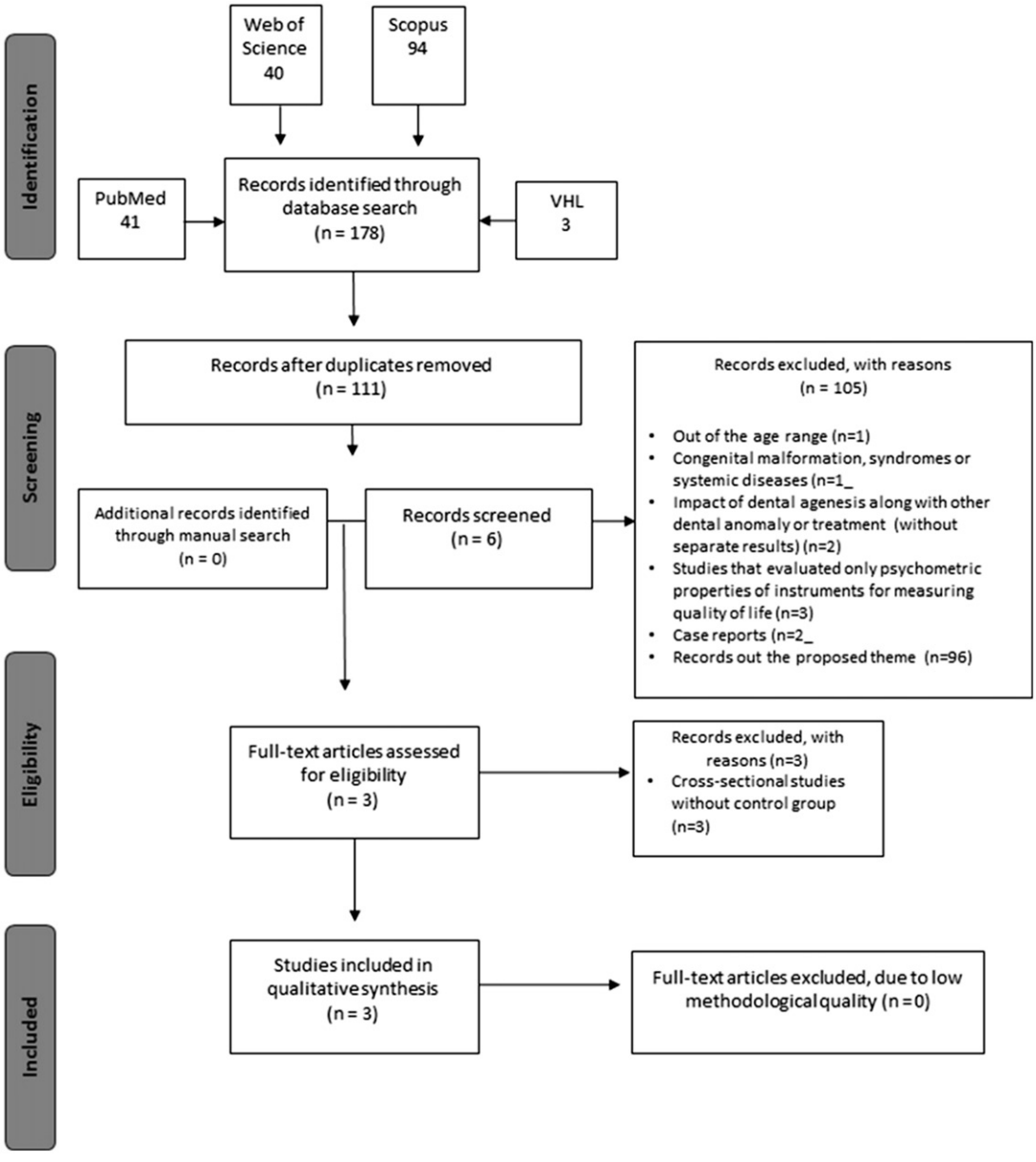


**Figure 1.** Flowchart describing the articles search process and selection based on the Preferred Reference Items for Systematic Reviews and Meta-Analyses (PRISMA).

Table 2. Evaluation of methodological quality and risk of bias according to Fowkes and Fulton [14].

| Questions | Items | | Laing et al. [17] | Kotecha et al. [18] | Hvaring et al. [19] |
|---|---|---|---|---|---|
| Study design appropriate to objectives? | Objective | Common design | | | |
| | Prevalence | Cross-sectional | NA | NA | NA |
| | Prognosis | Cohort | NA | NA | NA |
| | Treatment | Controlled trial | NA | NA | NA |
| | Cause | Cohort, case-control, Cross-sectional | 0 | 0 | 0 |
| Study sample representative? | Source of sample | | 0 | 0 | 0 |
| | Sampling method | | NA | NA | NA |
| | Sample size | | 0 | 0 | 0 |
| | Entry criteria/exclusions | | 0 | 0 | 0 |
| | Non-respondents | | NA | NA | NA |
| Control group acceptable? | Definition of controls | | + | 0 | + |
| | Source of controls | | 0 | 0 | 0 |
| | Matching/randomisation | | NA | NA | NA |
| | Comparable characteristics | | 0 | 0 | 0 |
| Quality of measurements and outcomes? | Validity | | 0 | + | 0 |
| | Reproducibility | | + | 0 | 0 |
| | Blindness | | NA | NA | NA |
| | Quality control | | 0 | 0 | 0 |
| Completeness? | Compliance | | NA | NA | NA |
| | Drop outs | | NA | NA | NA |
| | Deaths | | NA | NA | NA |
| | Missing data | | NA | NA | NA |
| Distorting influences? | Extraneous treatments | | NA | NA | NA |
| | Contamination | | NA | NA | NA |
| | Changes over time | | NA | NA | NA |
| | Confounding factors | | + | 0 | 0 |
| | Distortion reduced by analysis | | 0 | 0 | 0 |
| Summary of Issues | Bias – The results are wrongly biased in a certain direction? (Biased) | | Yes | Yes | Yes |
| | Confounding – There are serious confounders or other influences that distort the results? | | No | No | No |
| | Chance – The results occurred by chance? | | No | No | No |
| Scientific evidence | Methodological quality | | M | M | M |

NA = Not applicable; + = Minor problem; ++ = Major problem; 0 = no problem.
Scientific evidence: H: High; M: Moderate; L: Low.

six articles, three additional selected studies were excluded [11,15,16].

The final sample for this systematic review involved three eligible articles, to apply the quality assessment [17–19]. Based on the checklist that assess the methodological quality and the risk of bias by Fowkes and Fulton [14], all studies were of moderate methodological quality and present risk of bias as described in Table 2.

Two of the studies were conducted in the UK [17,18] and one in Norway [19]. The UK studies measured the outcome using the $CPQ_{11-14}$ instrument [17,18] and the Norwegian study [19] used the OIDP and OIDP CS instruments. The participants' ages ranged from 10 to 17 years old. There was no significant difference between female and male gender in the studies [17–19] and all studies used a case-control design (Table 3).

The impact of DA on OHRQoL, in the study by Laing et al. [17], no statistically significant difference reported in the total scores of the questionnaire between the case and control groups ($p = .566$). However, the group with DA had more difficulty chewing when the deciduous teeth associated with the missing permanent teeth had been exfoliated. The difficulty of chewing was associated with the severity of DA ($p = .030$) (Table 3).

Kotecha et al. [18] comparing the $CPQ_{11-14}$ scores in individuals with and without DA, there was a greater impact in the agenesis group with a statistical difference in all domains ($p = .001$). However, there was no statistical difference in $CPQ_{11-14}$ scores regarding the degree of severity dental agenesis. There was a moderate correlation between the quality of life of the parents and the child in the P-CPQ instrument (rho =50.46, $p = .001$) (Table 3).

In the Norwegian study [19], there was no statistical difference between the two groups in the OIDP overall score. However, there was a statistical difference in the CS OIDP scale between groups with and without DA on problems related to quality of life, severity (effect size 0.8, $p < .01$), anterior localization (effect size 0.7, $p < .01$) and maxillary dental agenesis (effect size 0.9, $p < .01$) (Table 3).

## Discussion

The psychosocial impact of DA in children, adolescents and young adults has received little attention in the scientific literature, specifically in children and young adults. The articles selected in this systematic review presented adolescents as samples. There is a need to increase the research in this area, to have better understanding of the perception of the consequences of this dental anomaly on the quality of life, and its effect on the daily life of those affected. Additionally, knowing the patient's expectations can contribute to the decision regarding the course of the treatment. To date, this is the first systematic review that has evaluated such an effect. The evaluation of the methodological quality of the selected studies allowed an accurate analysis and extrapolation of the findings.

**Table 3.** Synthesis of data from selected studies.

| Author, year (Country) | Study design | Groups evaluated | Sample size | | Sample age | Age range | Gender | | Instrument applied | Outcomes |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | Case group | Control group | | | Males | Females | | |
| Laing et al. 2010 (United Kingdom) [17] | Case-control | Dental agenesis | 62 | 61 | 11–16 years old | 13.6 (SD 1.6) | 61 | 62 | $CPQ_{11-14}$ | - No statistically significant difference in CPQ scores (overall or at domain level) or visual analog scores when comparing the case and control group.<br>- Differences were found in the group with dental agenesis in relation to some functional abilities compared to other patients with malocclusion characteristics. |
| Kotecha et al. 2013 (United Kingdom) [18] | Case-control | Mild agenesis (<6 agenesis) and severe agenesis (≥6 agenesis) | 86 | 30 | 11–14 years old | 12.5 (SD 1.0) | 49 | 67 | $CPQ_{11-14}$ | - Statistically significant difference was observed in the oral symptoms, functional limitations, and in the social and emotional well-being in the case group.<br>- Considering the number of missing teeth and the quality of life score, no statistical difference was observed.<br>- Other variables such as gender, socioeconomic status, the local of agenesis or the presence of retained primary teeth, did not influence the CPQ score. |
| | | | | | | | | | P-CPQ | - Moderate correlation between parental and child reported quality of life. |
| Hvaring et al. 2014 (Norway) [19] | Case-control | Dental agenesis | 62 | 101 | 10–17 years old | 12.9 (SD 1.5) | 80 | 83 | CS OIDP | - It was observed that the CS OIDP discriminated strongly between the case group and the control group regarding problems with emotional status, showing teeth, social contact, speaking and carrying out work.<br>- The impact on OHRQoL was related to the severity and the upper anterior localization of dental agenesis. |
| | | | | | | | | | OIDP | - The generic OIDP instrument did not detect statistically significant with respect to the overall scores. |

$CPQ_{11-14}$: Child Perceptions Questionnaire; P-CPQ: Parental-Caregiver Perceptions Questionnaire; CS OIDP: Condition specific Oral Impact on Daily Performance; OIDP: Oral Impact on Daily Performance.

Of the articles selected in this systematic review, only Kotecha et al. [18] found a more significant impact in the adolescent agenesis group, with a statistical difference in all domains. While Hvaring et al. [19] did not find statistical difference between case and control group but emphasized the impact on OHRQoL in cases of severe DA. These may be explained by that agenesis of lateral incisors being common in mild agenesis cases, resulting in a significant impact on OHRQoL since it is an aesthetic area [18]. However, Laing et al. [17], there was no evidence of any statistically significant relationships between the total CPQ scores of the DA group and in any of the independent variable (age, gender, total absolute DA, total relative DA, total absolute DA in the maxillary middle sextant and total relative DA).

The selected studies [17–19] used valid and reliable instruments to evaluate on OHRQoL: $CPQ_{11-14}$, CS OIDP and OIDP. The original version of $CPQ_{11-14}$ evaluated its psychometric properties with orthodontic and pediatric dentistry clinic in patients from 11 to 14 years old [20]. Considering the place of sample, Laing et al. [17] and Kotecha et al. [18] applied the $CPQ_{11-14}$ properly. In the study of Laing et al. [17], the participants were recruited from new patient orthodontic clinics in a teaching hospital, and Kotecha et al. [18] recruited from multidisciplinary clinics in a dental hospital.

Based on the age range, Kotecha et al. [18] applied the $CPQ_{11-14}$ properly, however, Laing et al. [17] used $CPQ_{11-14}$ in sample for ages 11–16 years. Laing et al. [17] should have tested the psychometric properties of $CPQ_{11-14}$ to confirm if this instrument was valid and reliable for this age range. According to Piassi et al. [21] it is important to test an instrument using the same sample that it was applied because the instrument may require adjustments for a particular group. In this concept, Hvaring et al. [19] used the adult version of OIDP and it was considered appropriate because the sample had a high number of adolescents (with ages up to 17 years) and the child version has been used between 6 and 13 years. They reported that the instrument used was translated into certified Norwegian language. And they also evaluated internal consistency considering the satisfactory generic OIDP for the studied age group and any of the items that had to be deleted.

From these quality assessments, the score demonstrated that the included studies had a moderate methodological quality and risk of bias. Some methodological aspects may have influenced the studies. In Laing et al. [17] and Kotecha et al. [18], the participants were instructed to complete the questionnaire on their own, without communication and without the assistance of a parent or guardian. However, in the study by Laing et al. [17], those who did not have enough time on the day they took the $CPQ_{11-14}$ and VAS home for the child to complete. In such cases, the participants may have been influenced or received help from a parent or guardian, even though they were aware that they could not help their children. In the study by Hvaring et al. [19], the participants completed a supervised self-administered questionnaire.

The final number of studies included in this systematic review was not the reason for not conducting meta-analysis, since according to Valentine et al. [22], meta-analysis can be done with at least two studies. However, it was the methodological aspect that made it difficult to compare the studies and consequently carry out a meta-analysis with different cut-off points that classified the impact on OHRQoL. Kotecha et al. [18] utilized the 16-item short-form, while Laing et al. [17] used the full form of $CPQ_{11-14}$ with the 37 questions while Hvaring et al. [19] used another quality of life instrument (CS OIDP/OIDP).

The probability of risk of bias in this systematic review was low, since the search was performed either manually or using considerable databases for all bibliographic references of the selected articles. We also reached to the grey literature to identify unpublished and ongoing studies and considered other languages. We also used common MESH terms and keywords from articles published in the area in order to minimize sources of inconsistency and the possibility of not finding potentially eligible studies.

The quality assessment detected other potential limitations, such as the definition of controls and the validity of the quality of life instruments. It is important to standardize the methods of analysis OHRQoL of children, adolescents and young adults to obtain evidence-based outcomes.

This systematic review provided the first evidence on the significant impact of DA on OHRQoL. It is suggested that DA does not generate impact on OHRQoL of adolescents. But this should be interpreted with caution, since only few studies evaluated this special condition.

It is of important relevance, because it can improve the appreciation of the psychosocial changes and their impact on OHRQoL and that they should be considered in the therapeutic approach of this group.

## Conclusions

Insufficient evidence is available to conclude whether DA affects OHRQoL of children, adolescents and young adults. We recommend further studies on this theme clarify these questions.

## Disclosure statement

No potential conflict of interest was reported by the authors.

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