

# The impact of cleft lip and palate on the quality of life of young children: A scoping review

Muhammad Safwan Yusof, Bsc, Hasherah Mohd Ibrahim, PhD

Universiti Kebangsaan Malaysia, Faculty of Health Science, Jalan Raja Muda Abdul Aziz, Kuala Lumpur, Malaysia

## ABSTRACT

**Introduction:** Cleft lip and palate (CL/P) are among the most common congenital abnormalities. The purpose of the present study was to review the literature relating to the quality of life (QoL) in young patients with cleft lip and/or palate (CL/P) and to identify the specific aspect of QoL in young patients with CL/P that is mostly affected. Other associated variables within studies that may have an impact on QoL were also identified.

**Materials and Methods:** Systematic searches of PubMed, Scopus and Web of Science databases were conducted. Independent reviewers screened the title, abstract and full texts according to predetermined inclusion and exclusion criteria. Articles published in English from January 2012 to March 2022 reporting the QoL of non-syndromic young patients aged 7–18 years with CL/P were included. Review articles and articles reporting the psychological adjustment of parents or other family members with CL/P were excluded.

**Results:** 975 publications were identified, of which 20 studies met our inclusion criteria. The majority of studies reported that the CL/P condition has a negative impact on the QoL. Psychological health, functional well-being, social-emotional well-being and school environment are domains that are affected. Compared with typically developing young patients, those with CL/P had lower QoL scores even though QoL was assessed using different instruments across studies. The impact of CL/P on overall QoL scores varied by age but not gender or cleft type.

**Conclusion:** Our reviews had shown the presence of CL/P negatively affects the QoL of young patients. Psychological health is the most affected QoL domain. Understanding the impacted domain will help in planning and delivering better health care for individuals with CL/P and reducing the stigma commonly associated with CL/P. Future studies should target intervention on psychological health and consider resilience factors towards positive adjustment.

## KEYWORDS:

*Quality of life, cleft lip, cleft palate, congenital, children, adolescents*

## INTRODUCTION

Cleft lip and palate (CL/P) are among the most common congenital abnormalities, with an overall worldwide prevalence of 1 per 1,000 births.<sup>1</sup> The management of CL/P is

long-term, beginning from birth and continuing into early adulthood. While a child born with CL/P faces a visible facial disfigurement, they also encounter other issues related to the cleft such as feeding, hearing, speech and language difficulties that compromise their overall ability to communicate effectively. Young patients with CL/P are at greater risk of developing psychological problems due to the various issues associated with having cleft.<sup>2,3</sup> Some contributing factors include parental stress and worry,<sup>4</sup> difficulties coping with academic demands,<sup>5</sup> and being teased or bullied due to having visible differences and speech and hearing difficulties.<sup>6</sup>

The distress may manifest itself through various psychological and psychosocial problems such as anxiety and depression, emotional and behavioural issues, poor social skills, social withdrawal, poor self-concept and lower self-esteem. These problems become more apparent at schoolage as physical aesthetic and speech quality becomes the key factors to successful social interaction and acceptance among peers.<sup>7</sup> Young patients with CL/P, especially the ones with visible facial asymmetries and scarring, may face social rejection, experience more events of teasing and bullying at school, and have a lower quality of life (QoL) score when compared to those with less visible cleft features, as seen in cases of cleft palate only.<sup>5,8</sup> Unfortunately, being teased or bullied has been linked to poorer psychosocial adjustments; increasing the likelihood of developing psychiatric-related issues later in life.<sup>9,10</sup>

Previous studies have shown that young patients with CL/P have a poor health-related QoL compared to unaffected peers,<sup>11-14</sup> albeit not always consistently.<sup>15,16</sup> These inconsistent findings may be attributed to factors such as sample size, place of study and the involvement of multidisciplinary care and support from a psychological team or lack thereof.<sup>17-20</sup> For example, Tannure et al.<sup>16</sup> showed that delivering psychological and surgical intervention during early childhood improved the QoL of both patients and their caregivers.

In the past decade, two systematic reviews have been conducted by Klassen et al.<sup>21</sup> and Herkrath et al.<sup>22</sup> on the QoL of young patients with CL/P. Klassen et al.<sup>21</sup> identified health concepts and determinants of QoL in individuals with CL/P and outlined a conceptual framework of QoL that includes physical, psychological and social health. This review found that while several domains such as physical health, self-esteem, psychological distress and peer relation are well-researched among affected individuals, other areas such as

*This article was accepted: 29 January 2023*

*Corresponding Author: Hasherah Mohd Ibrahim*

*Email: hasherah@ukm.edu.my*

family function, social function, social support and school function remained poorly studied.<sup>21</sup> Klassen et al.<sup>21</sup> also identified several instruments used to assess QoL in young patients with CL/P, such as the Youth Quality of Life Instrument-Craniofacial Surgery (YQoL-CS) and Child Oral Health Quality of Life Questionnaire (COHQoL). Importantly, they noted that these questionnaires focussed broadly on craniofacial conditions and did not include specific concerns of young patients with CL/P conditions.<sup>21</sup> On the other hand, Herkrath et al.<sup>22</sup> focussed on the QoL of young patients with nonsyndromic CL/P and reported that CL/P negatively affects the QoL in at least one domain with emotional and functional well-being as the most affected domains and social dimension as the least affected.

Identifying predictors of QoL and associated risks factor is essential in planning and delivering better health care for individuals with CL/P and reducing the stigma commonly associated with CL/P.<sup>23,24</sup> However, while the earlier reviews by Klassen et al.<sup>21</sup> and De Queiroz Herkrath et al.<sup>25</sup> made a significant contribution towards this goal, neither reported the impact of CL/P on QoL by age, gender or type of cleft. Therefore, the purpose of this review is to (1) systematically review the literature relating to the QoL in young patients with cleft lip and/or palate CL/P and (2) to identify the specific aspect of QoL in young patients with CL/P such as age, gender and cleft types that may have an impact on specific QoL domains (oral health, functional well-being and social-emotional) that is mostly affected.

## MATERIALS AND METHODS

This scoping review was conducted based on the five-stage methodological framework proposed by Arksey and O'Malley.<sup>26</sup> The five stages include (1) identifying the research questions, (2) identifying relevant studies, (3) study selection, (4) charting the data and (5) collating, summarising and reporting the results.

### Inclusion Criteria

Original articles reporting the QoL of patients aged 7–18 years with CL/P were included to assess the impact of CL/P conditions on school-aged patients specifically. Throughout the manuscript, the terms young patient with CL/P were used to avoid confusion with children and adolescent-specific definitions in the result later on. Convention on the Rights of the Child (CRC), defined a child as “every human being below the age of eighteen years unless under the law applicable to the child, majority is attained earlier”.<sup>27</sup> However in this review paper, we are interested in children aged 7–18 years old. Relevant studies published in English from January 2012 to March 2022 utilising quantitative, qualitative or mixed-method modalities were considered. In addition, studies with any reporting modality, including self-reports, parent reports and third-party reports (such as those obtained via clinicians, laypersons and teachers), were included.

### Exclusion Criteria

Literature reviews, systematic reviews, meta-analyses, summary articles, book chapters, case studies, letters, comments, editorials and unpublished dissertations published during the search period were excluded. Articles relating to other physical disfigurements of cleft lip and

palate were excluded. Also, articles reporting the psychological adjustment of parents or other family members with CL/P were excluded as this study aimed to examine only the QoL of children with CL/P. Articles reporting ‘late presentation’ for cleft repair in children, adolescents, young adults or adults were excluded since the findings are not equivalent to routine treatment. Finally, articles that did not differentiate the results of nonsyndromic CL/P and syndromic children were excluded.

### Search Strategy

Three online databases (PubMed, Scopus and Web of Sciences) were searched in March 2022 to identify potentially relevant articles. The search string used was (“cleft lip palate” OR “cleft lip” OR “cleft palate”) AND (“quality of life”) AND (children OR teenager OR youth OR adolescent). No articles were recovered from grey literature.

### Study Selection Process

During the study selection process, inclusion and exclusion criteria were used to select the study in line with scoping review method. Abstracts were obtained for all the studies identified during electronic searches. Two reviewers (SY and HM) independently screened the title, abstracts and full-text copies to eliminate articles that failed to meet eligibility criteria.

### Charting the Data

A data extraction form was created using commercial spreadsheet software (Microsoft Excel™365, Microsoft, Inc., Redmond, WA, USA) by SY to summarize the data. Only articles meeting the inclusion criteria were included in reviews. The reviewers discuss whether the data being extracted answered research questions. Following revisions, the final data charted were: author (s), age range, sample size, types of cleft, instruments used, informant type, consensus, determinant, reported negative influence in CL/P and associated factors.

## RESULTS

The electronic search generates 975 results. After removing duplicates, 532 unique articles were identified. Title and abstract screening resulted in the exclusion of 455 articles. The full texts of 77 articles were retrieved and another 57 articles were excluded after full-text screening for not meeting the inclusion criteria. Finally, 20 articles were included in this scoping review (Table I). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guideline<sup>28</sup> was adapted for use in this scoping review (Figure 1).

Due to different instruments being used to access QoL across studies, a narrative approach was used to report the findings of the included studies. The results and conclusion of this review are presented by highlighting any statistically significant findings reported from original articles.

### Respondents

The study population in 11 out of the 20 studies (55%) included in this review comprised young patients with CL/P (mean age=12.8 years),<sup>12,29-38</sup> while nine studies (45%) involved child-parent dyad.<sup>13,18-20,39-43</sup> Seven studies reported similar findings or no statistically significant differences

Table 1: Characteristics of the studies included in the review

No	Author	Cleft/control (N)	Age range; types of cleft	QOL instruments, informant, correlation/consensus	Determinants	Impacted domain
1	Oka (29) <sup>a</sup>	69	11-18; CL, CP	YQOL-FD, children	Age; sex; cleft types	<ul style="list-style-type: none"> <li>Negative consequence and stigma</li> <li>Age (significant)</li> <li>Caregiver and children rating (overall score and oral symptoms)</li> <li>Peers (overall score)</li> </ul>
2	Ali (39) <sup>a</sup>	70	8-16; CLP	COHIP, Children, and caregiver, significant differences	Children and parents	
3	Ali (30) <sup>a</sup>	75/150	8-16; CLP	COHIP, children	Age; gender; peers, cleft types	
4	Fowler (40) <sup>a</sup>	174	8-13 CP, CLP	CPQ & Parent Version (P-CPQ); children and parents; good correlation (r= 0.97)	Sex, Ethnicity, cleft phenotype	<ul style="list-style-type: none"> <li>Not significance on total score</li> <li>Cleft phenotype (not significance – CLP had a higher score compared to CP)</li> <li>Ethnicity (significance- the pacific island had a higher score)</li> <li>Cleft phenotype</li> <li>Self-esteem did not differ in significance for both groups except for behavioral conduct and scholastic competence</li> <li>higher self-esteem in cleft groups</li> <li>Cleft types</li> <li>Gender</li> <li>Age</li> </ul>
5	Aleksieva (31) <sup>a</sup>	91/790	CLP: 12.74 ± 1.86 Control: 12.76 ± 1.26; CL, CP, CLP	CPQ, children	Cleft and control, self-esteem	<ul style="list-style-type: none"> <li>Not significant when compared between age and gender</li> <li>Gender (female lower score in Bodily pain, vitality, and mental health)</li> <li>Cleft type (CL and CP lower score of HRQOL than CLP in domain: limitation)</li> </ul>
6	Lin (13) <sup>a</sup>	120	8-15; CL, CP, CLP	COHIP, children and parents, weak-moderate correlation	Age, gender; cleft types, parents	
7	Crepaldi (32)	57	14-17; CL, CP, CLP	SF-36, children	CLP types, age, gender	
8	Nolte (41)	170	8-18; CL, CP, CLP	COHIP, children and parents, a significant difference in oral symptoms and functional well-being only	Gender, parents, cleft types	
9	Nagappan (33) <sup>a</sup>	80/80	8-16; CL, CP,	COHIP, children	Cleft and control	<ul style="list-style-type: none"> <li>Functional well-being</li> <li>Social/ emotional well-being,</li> <li>School environment</li> </ul>
10	Abebe (19)	41	12.37 ± 2.5; CL, CP	COHIP, children and parents, strong internal reliability	Rating parents and children	
11	Ajami (34) <sup>a</sup>	50/50	8-15; CLP	COHIP, children	gender, control, type-D	<ul style="list-style-type: none"> <li>Gender (emotional well-being), Age (oral symptoms)</li> </ul>
12	Broder (42) <sup>a</sup>	1196	7.5-18.5; CP, CLP	COHIP, children and caregiver	Gender, Cleft type, insurance	<ul style="list-style-type: none"> <li>Surgical recommendation</li> <li>Gender</li> <li>Surgery</li> <li>Insurance</li> <li>Visibility of cleft</li> <li>Type of cleft</li> <li>Age (socio-emotional well-being)</li> <li>Type of cleft (functional well-being)</li> </ul>
13	Agnew (18) <sup>a</sup>	222	7-18; CL, CP	COHIP-SF, children and parents, strong correlation	Age; agreement; types of cleft, private insurance	
14	Aravena (35) <sup>a</sup>	48/96	8-15; CL, CP, CLP	COHIP, children	Control	<ul style="list-style-type: none"> <li>CLP and control (functional well-being, school environment, self-image)</li> </ul>

cont..... pg 253

cont from..... pg 252

Table I: Characteristics of the studies included in the review

No	Author	Cleft/control (N)	Age range; types of cleft	QOL instruments, informant, correlation/consensus	Determinants	Impacted domain
15	Vuletic (36) <sup>a</sup>	73/70	11-18; CL, CP, CLP	QLACA, children	Control, gender, age	<ul style="list-style-type: none"> <li>Relationship with parents,</li> <li>Success</li> <li>Society</li> <li>Appearances</li> <li>Function</li> <li>Compare control (total score, functional limitations, emotional well-being, social well-being)</li> <li>Patient and parent (self-image)</li> <li>Cleft type (total score and functional well-being)</li> </ul>
16	Kortelainen (12) <sup>a</sup>	51/82	11-14; CLP	CPQ, children	control, age, gender	
17	Konan (20)	140	8-15; CLA, CLP	COHIP, children and parents, no significance between parent and children rating	Age, gender, parents' reports	
18	Broder (37) <sup>a</sup>	1200	7-18; CP, CLP	COHIP, children	Gender, age	<ul style="list-style-type: none"> <li>Surgical recommendation</li> <li>Oral health</li> <li>Functional well-being</li> <li>Socio-emotional well-being, school/environment</li> <li>Oral health,</li> <li>Functional well-being</li> <li>Social emotional well-being</li> <li>Emotional well-being on gender</li> </ul>
19	Ward (43) <sup>a</sup>	75/75	8-18; CL, CP, CLP	COHIP, children and parents, no significant difference	Age, caregiver	
20	Eslami (38) <sup>a</sup>	50	8-15; CLP	COHIP, children	Age, gender, type of cleft	

<sup>a</sup>Reported negative influence of CL/P on QoL.

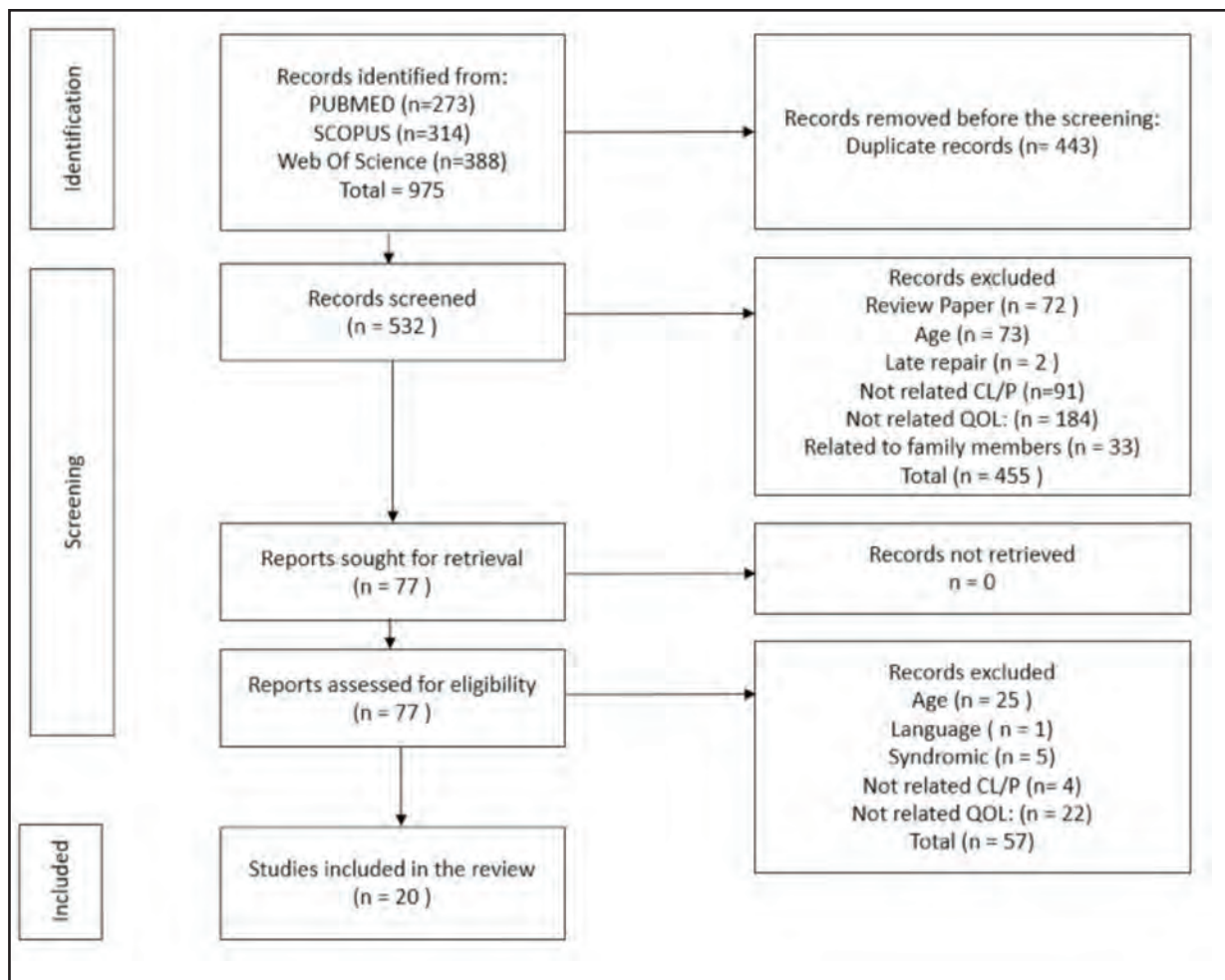


Fig. 1: Flow of the literature search and the selection process

between self-reported or parent-reported QoL of young patients with CL/P.<sup>18-20,40-43</sup> However, two studies reported weak correlation or statistical significance between young patients and caregivers.<sup>13,39</sup> These findings indicate that young patients with CL/P are capable of reporting their QoL.

*QoL of Young Patients with CL/P*

There was some variation in the reported QoL of young patients with CL/P in the included studies. Sixteen studies reported the negative impact of CL/P on QoL,<sup>12,13,18,29-31,33-40,42</sup> while four articles reported a null association between CL/P and QoL.<sup>19,20,32,41</sup> Three of these four studies evaluated QoL using Child Oral Health Impact Profile (COHIP) as their instrument and reported an overall COHIP score of >100 among young patients with CL/P, indicating good QoL.<sup>19,20 41</sup>

Nine studies compared the QoL between young patients with CL/P with unaffected control or peer groups,<sup>12,30,31,33-36, 43,44</sup> of which six reported statistically significant differences in the overall QoL score of young patients with CL/P compared to their peers.<sup>12,30,31,33,36,43</sup> As expected, young patients with CL/P had lower QoL scores than unaffected peers even though different instruments were used to evaluate QoL, such as COHIP, Child-Oral Impacts in Daily Performance (Child-OIDP), CPQ and QLACA.

When looking at specific QoL domains, young patients with CL/P and peers show statistically significant ( $p<0.001$ ) differences in functional, social-emotional well-being and school environment.<sup>33,43</sup> Aleksieva<sup>31</sup> reported that all domains were significantly different between young patients with CL/P and their peers (total CPQ score, social, functional, emotional) except oral symptoms. Ward<sup>43</sup> demonstrated a significant interaction between age and social-emotional well-being in 15 to 18 years old compared to the 8 to 14 years age group. Similarly, Aravena et al.<sup>35</sup> showed a statistically significant difference ( $p<0.05$ ) in functional well-being, school environment and self-images between control and young patients with CL/P; however, the overall QoL score on COHIP was not statistically significant between the two groups.

*QoL of Young Patients with CL/P by Age*

Nine studies used age as a determinant in their analysis. However, since young patients is a wide age range (7–18 years) were included, we dichotomised the study participants into 7–12 years old (children) and 13–18 years old (adolescents). When the age range limit exceeded these categories' boundaries, the group was defined by the mean age.



The effect of CL/P among children and adolescents has been inconsistently reported: Five studies reported a low overall QoL score but no statistically significant difference between children and adolescents.<sup>12,30,36,38,43</sup> For example, Ali et al.<sup>30</sup> and Eslami et al.<sup>38</sup> reported overall COHIP scores of  $87.83 \pm 20.61$  and  $87.27 \pm 23.49$  among children and  $91.42 \pm 19.25$  and  $96.46 \pm 28.92$  among adolescents, respectively. Konan et al.<sup>20</sup> reported high overall COHIP scores among children and adolescents but no statistically significant between-group differences. The remaining three studies reported poorer overall QoL scores among adolescents than children.<sup>13,18,29</sup>

Regarding domain-specific differences, Agnew et al.<sup>18</sup> reported that adolescents scored lower on overall and socio-emotional domains, while Lin et al.<sup>13</sup> reported that adolescents scored lower on overall, functional and emotional domains. However, Oka et al.<sup>29</sup> observed that adolescents reported lower QoL scores in all domains (stigma, negative consequence, negative self-image, positive consequence) except the coping domain.

#### *QoL of young patients with CL/P by gender*

Most studies did not find any significant difference in the overall QoL score by gender.<sup>12,13,18,20,29,30,32,34,36,38,41,42</sup> However, there were significant differences in specific domains. For example, three studies reported significant differences in the emotional well-being domain.<sup>13,34,38</sup> In a study by Broder et al.<sup>42</sup>, female participants had lower self-rated emotional well-being and overall COHIP scores than male participants. Furthermore, Crepaldi et al.<sup>32</sup> reported that females scored lower in bodily pain, vitality and mental health domains. In contrast, Nolte et al.<sup>41</sup> reported that females scored significantly higher (higher QoL) on functional well-being and in the school environment.

#### *QoL of young patients with CL/P by type of cleft*

Nine studies reported no significant differences in QoL between cleft types<sup>18,29-32,38,40-42</sup> except Lin et al.<sup>13</sup>, who reported significant differences in overall score between cleft lip (CL), cleft palate (CP) and cleft lip and palate (CLP). However, some studies reported domain-specific differences in QoL between cleft phenotypes. Six studies reported poorer QoL of young patients with CLP but were not statistically significant compared to CL and CP.<sup>30,31,38,40-42</sup> In contrast, Lin et al.<sup>13</sup> and Crepaldi et al.<sup>32</sup> reported that young patients with CL had lower QoL scores than patients with CLP.

Crepaldi et al.<sup>32</sup> also reported that young patients with CL and CP had statistically lower scores in emotional and mental health domains than those with CLP. Similarly, Agnew et al.<sup>18</sup> and Nolte et al.<sup>41</sup> reported poor functional well-being among young patients with CLP. In contrast, Aleksieva et al.<sup>31</sup> reported significant differences in oral symptoms and functional restriction in young patients with CLP. These inconsistent findings may be due to the timing of receiving treatment and methodological differences, such as the distribution of cleft types between studies.<sup>30,32</sup> Nevertheless, some studies in this review did not analyse the types of cleft separately, which may result in bias.

## DISCUSSION

This scoping review aimed to identify the impact of CL/P on the QoL of young patients. A total of 20 studies were reviewed, all of which evaluated the QoL of nonsyndromic participants aged 7–18 years with CL/P using quantitative methods. In line with previous studies,<sup>19</sup> oral health-related QoL was commonly used to assess the outcome of multidisciplinary cleft care. The findings from this scoping review confirm that the CL/P condition affects the overall QoL scores of young patients with CL/P compared to their typically developing peers.<sup>12,30,31,33,36,43</sup> These findings might be explained by the fact that young patients with CL/P have more challenges at school, such as social interaction, having to undergo cleft-related treatment and aesthetic-related concerns, compared to unaffected peers.<sup>5,45</sup> However, three studies that use COHIP indicated relatively positive QoL (mean score = 120–155.56) in young patients with CL/P.<sup>19,20,41</sup> Three studies revealed positive outcomes because multidisciplinary care received as all three studies recruited participants with CL/P attended by multidisciplinary care teams from university hospitals, which may have resulted in favourable QoL scores.

Although different instruments were used to measure QoL, these instruments have been found to demonstrate reliability and validity values.<sup>36,46-50</sup> The main difference between instruments is the constructs measure. For example, COHIP measures oral health, functional well-being, social-emotional, school environment and self-image. Meanwhile, the YQOL-FD evaluate stigma, negative self-image, positive consequence, negative consequence and coping. CPQ measure oral symptoms, functional limitation, emotional well-being and social well-being.

We also reviewed the QoL of young participants affected by CL/P by age, gender and cleft type. The age-specific effects of CL/P on participants' QoL were heterogeneous. Three out of nine studies that used age as a determiner reported poorer QoL among adolescents (13–18 years old) with CL/P than children (7–12 years old) with CL/P,<sup>13,18,29</sup> especially in social-emotional well-being. These findings may be because adolescents are more concerned regarding their facial appearances as they need to cope with the facial difference in addition to typical adolescent concerns regarding appearances.<sup>18,51</sup> However, five studies reported low overall scores but no significant difference in the QoL between children and adolescents.<sup>12,30,36,38,43</sup> In contrast, Konan et al.<sup>20</sup> reported a numerically high overall QoL score but no significant difference between children and adolescents. There may be at least three reasons for this finding; (1) the small age range encompassing the two groups<sup>20,30</sup> (2) children with CL/P were as aware of their condition and had similar experiences as adolescents,<sup>38</sup> or (3) the studies with inadequately powered to detect age-specific differences. For instance, studies by Lin et al.<sup>13</sup> and Agnew et al.<sup>18</sup>, which reported poorer QoL among adolescents compared to children, had larger sample sizes ( $n > 120$ ) compared to the five studies ( $n = 51-75$ ) that found no statistical difference between children and adolescent.

Regarding gender, although there was no significant between-group difference in the overall QoL scores, significant differences in domain-specific QoL scores were

noted between males and females. For example, emotional well-being was the most affected domain in females compared to males,<sup>13,34,37,38</sup> in line with earlier studies indicating that females tend to be more self-conscious and place greater importance on their appearances than males.<sup>21,52,53</sup> Similarly, nine studies showed no significant differences in the overall QoL scores by cleft types.<sup>18,29-32,38,40-42</sup> We also found that the QoL of young patients with cleft lip and palate is poorer, albeit not significantly, compared to patients with cleft palate.<sup>30,31,38,40-42</sup> These findings are similar to an earlier review by Hunt et al.<sup>54</sup> that reported that the type of cleft and its severity appear to have little impact on the individual's overall psychosocial functioning. It is plausible, though, that patients with visible defects (CL or CLP) may be more dissatisfied with their appearance than those without a visible cleft defect. Accordingly, Crepaldi et al.<sup>32</sup> and Lin et al.<sup>13</sup> reported poorer QoL in patients with cleft palate than those with cleft lip and palate. One of the reasons for this discrepancy is methodological differences, such as the unequal distribution of cleft types due to the unbalanced structure of participants with CL/P.<sup>13</sup> Secondly, those with more complex clefts may emphasise the rehabilitative process, such as facial appearances, while those with less complex clefts may consider functional aspects, such as speech.

Overall, psychological health was the most affected QoL domain in young patients with CL/P. In addition, other QoL dimensions include functional well-being (impact on the ability to carry out a specific task, e.g., speaking clearly, chewing), social-emotional well-being (implications for peer interaction and mood states) and school environment (impact on functions associated with school environment) seems to be negatively affected in young patients with CL/P. In contrast, oral health (impact on oral symptoms, e.g., pain, spots on teeth) and physical health were the least affected QoL domains.<sup>32</sup> This finding is similar to those reported in earlier reviews by Herkrath et al.<sup>22</sup> and Hunt et al.<sup>54</sup>, which found that emotional and functional well-being are most affected in young patients with CL/P.

A plausible explanation for poor functional well-being is that young patients with CL/P have difficulty eating or speaking due to missing or rotated teeth. They may also have problems keeping their teeth clean and most children with CL/P have an orthodontic appliance which can further contribute to functional difficulties.<sup>30,43,55</sup> Meanwhile, challenges dealing with societal norms and expectations regarding facial appearances and communicative skills may severely affect the emotional well-being of young patients with CL/P. Furthermore, they may be more worried or anxious, experience teasing or bullying and be concerned about how others perceive them.<sup>18,43</sup> The school environment is another negatively impacted domain among young patients with CL/P, as also noted by Stock and Feragen.<sup>56</sup> For example, patients with CL/P may have otitis media; thus, they struggle at school, need to sit at the front of the class and may require more support, such as a hearing aid.<sup>57</sup> In addition, young patients with CL/P miss more school days than unaffected peers due to hospital appointments for cleft-related treatment.<sup>45</sup>

## LIMITATIONS

Although we conducted this scoping review based on the PRISMA statement and used a meticulous literature search strategy, we did not include grey literature or literature published in a non-English language, which may have inadvertently led to the exclusion of some relevant research. Also, we could not assess the impact of treatment duration or patient resilience on QoL outcomes of young patients with CL/P as these areas are poorly researched.

## CONCLUSION

The current review found that most studies report poor QoL outcomes in young patients with CL/P, especially in the psychological health, functional well-being, social-emotional well-being and school environment QoL domains. While different tools were used to measure QoL, the tools were generally giving consistent results with the outcomes and caregiver ratings. While QoL outcomes between children and adolescents with CL/P are inconsistently reported, the current evidence does not indicate exacerbated QoL outcomes by gender or cleft type. More studies investigating the QoL of young patients with CL/P with a larger sample size that can be representative of the population are warranted. Additionally, future studies should consider targeted prevention measures for helping young patients in the areas of psychological health, functional well-being, social-emotional well-being and school environment domains QoL domains. Resilience towards positive adjustment and the socio-economic status of young patients with CL/P should be considered as these factors may influence QoL outcomes. The World Health Organization has highlighted that assessing socio-economic characteristics is pertinent to understanding QoL outcomes.<sup>58-60</sup>

## CONFLICT OF INTEREST AND FUNDING

The authors declare no conflicts of interest. This study was funded by the Ministry of Higher Education, Malaysia (FRGS/1/2021/SSO/UKM/02/2)

## REFERENCES

1. Mossey PA, Modell B. Epidemiology of oral clefts 2012: an international perspective. *Cleft Lip Palate*. 2012; 16: 1-18.
2. Kappen IF, Bittermann GK, Stock NM, Mink van der Molen AB, Breugem CC, Swanenburg de Veye HF. Quality of life and patient satisfaction in adults treated for a cleft lip and palate: a qualitative analysis. *Cleft Palate-Craniofacial J* 2019; 56(9): 1171-80.
3. Ardouin K, Hare J, Stock NM. Emotional well-being in adults born with cleft lip and/or palate: a whole of life survey in the United Kingdom. *Cleft Palate-Craniofacial J* 2020; 57(7): 877-85.
4. Lockhart E. The mental health needs of children and adolescents with cleft lip and/or palate. *Clin Child Psychol Psychiatry*. 2003; 8(1): 7-16.
5. Glener AD, Allori AC, Shammam RL, Carlson AR, Pien IJ, Aylsworth AS, et al. A population-based exploration of the social implications associated with cleft lip and/or palate. *Plastic Reconstr Surg Glob Open*. 2017; 5(6).
6. Nicholls W, Selvey LA, Harper C, Persson M, Robinson S. The psychosocial impact of cleft in a Western Australian cohort across 3 age groups. *Cleft Palate-Craniofacial J*. 2019; 56(2): 210-21.

7. De Sousa A, Devare S, Ghanshani J. Psychological issues in cleft lip and cleft palate. *J Indian Assoc Pediatr Surg* 2009; 14(2): 55.
8. Feragen KB, Særvold TK, Aukner R, Stock NM. Speech, language, and reading in 10-year-olds with cleft: associations with teasing, satisfaction with speech, and psychological adjustment. *Cleft Palate-Craniofacial J* 2017; 54(2):1 53-65.
9. Hunt O, Burden D, Hepper P, Stevenson M, Johnston C. Self-reports of psychosocial functioning among children and young adults with cleft lip and palate. *Cleft Palate-Craniofacial J* 2006; 43(5): 598-605.
10. Vanderbilt D, Augustyn M. The effects of bullying. *Paediatr Child Health* 2010; 20(7): 315-20.
11. Alka Mariam M. Oral health related quality of life among cleft lip and palate patients: questionnaire: Ragas Dental College and Hospital, Chennai; 2020.
12. Kortelainen T, Tolvanen M, Luoto A, Ylikontiola LP, Sandor GK, Lahti S. Comparison of oral health-related quality of life among schoolchildren with and without cleft lip and/or palate. *Cleft Palate Craniofac J* 2016; 53(5): e172-6.
13. Lin J, Fang X, Ha P, Fu M, Wang H. Oral health-related quality of life in chinese children with orofacial cleft. *Cleft Palate Craniofac J* 2020; 57(8): 931-7.
14. Long RE, Wilson-Genderson M, Grayson BH, Flores R, Broder HL. Oral health-related quality of life and self-rated speech in children with existing fistulas in mid-childhood and adolescence. *Cleft Palate Craniofac J* 2016; 53(6): 664-9.
15. Sundell AL, Tornhage CJ, Marcusson A. A comparison of health-related quality of life in 5- and 10-year-old Swedish children with and without cleft lip and/or palate. *Int J Paediatr Dent* 2017; 27(4): 238-46.
16. Tannure PN, Soares FM, Kuchler EC, Motta LG, Costa MC, Granjeiro JM. Measuring the impact of quality of life of children treated for orofacial clefts: a case-control study. *J Clin Pediatr Dent* 2013; 37(4): 381-4.
17. Funder DC, Ozer DJ. Evaluating effect size in psychological research: sense and nonsense. *Adv Methods Practices Psychol Sci* 2019; 2(2): 156-68.
18. Agnew CM, Foster Page L, Hibbert S. Validity and reliability of the COHIP-SF in Australian children with orofacial cleft. *Int J Paediatr Dent* 2017;27(6):574-82.
19. Abebe ME, Deressa W, Oladugba V, Owais A, Hailu T, Abate F, et al. Oral health-related quality of life of children born with orofacial clefts in ethiopia and their parents. *Cleft Palate Craniofac J* 2018; 55(8): 1055665618760619.
20. Konan P, Manosudprasit M, Pisek P, Pisek A, Wangsrimgkol T. Oral health-related quality of life in children and young adolescent orthodontic cleft patients. *J Med Assoc Thai* 2015; 98 Suppl 7: S84-91.
21. Klassen AF, Tsangaris E, Forrest CR, Wong KW, Pusic AL, Cano SJ, et al. Quality of life of children treated for cleft lip and/or palate: a systematic review. *J Plast Reconstr Aesthet Surg* 2012; 65(5): 547-57.
22. Queiroz Herkrath AP, Herkrath FJ, Rebelo MA, Vettore MV. Measurement of health-related and oral health-related quality of life among individuals with nonsyndromic orofacial clefts: a systematic review and meta-analysis. *Cleft Palate Craniofac J* 2015; 52(2): 157-72.
23. Thompson JMD, Ayrey SL, Slykerman RF, Stone PR, Fowler PV. Quality of life using general population validated questionnaires in children with cleft lip and/or palate in New Zealand. *Cleft Palate Craniofac J* 2021; 58(6): 779-86.
24. Chung KY, Sorouri K, Wang L, Suryavanshi T, Fisher D. The impact of social stigma for children with cleft lip and/or palate in low-resource areas: a systematic review. *Plast Reconstr Surg Glob Open* 2019; 7(10): e2487.
25. De Queiroz Herkrath APC, Herkrath FJ, Rebelo MAB, Vettore MV. Measurement of health-related and oral health-related quality of life among individuals with nonsyndromic orofacial clefts: a systematic review and meta-analysis. *Cleft Palate-Craniofacial J* 2015; 52(2): 157-72.
26. Arksey H, O'Malley L. Scoping studies: towards a methodological framework. *Int J Soc Res Methodol* 2005; 8(1): 19-32.
27. Assembly UG. Convention on the Rights of the Child. United Nations, Treaty Series. 1989; 1577(3): 1-23.
28. Haddaway NRAUPCC, McGuinness LA. PRISMA2020: R package and ShinyApp for producing PRISMA 2020 compliant flow diagrams (Version 0.0.2). Zenodo; 2021.
29. Oka A, Tanikawa C, Isogai Y, Mihara K, Yamashiro T. Evaluation of facial appearance-related quality of life in young japanese patients with cleft lip and/or palate. *Cleft Palate Craniofac J* 2022; 59(4\_suppl2): S57-S64.
30. Ali MA, Nasir AF, Abass SK. Oral health-related quality of life among Sudanese children treated for cleft lip and palate. *Cleft Palate Craniofac J* 2021; 58(11): 1405-11.
31. Aleksieva A, Begnoni G, Verdonck A, Laenen A, Willems G, Cadenas de Llano-Perula M. Self-esteem and oral health-related quality of life within a cleft lip and/or palate population: a prospective cohort study. *Int J Environ Res Public Health* 2021; 18(11).
32. Crepaldi TA, Vitor LLR, Carrara CFC, Rios D, Cruvinel T, Almeida A, et al. Do cleft lip and palate types affect health-related quality of life of adolescents? *J Craniofac Surg* 2019; 30(3): 829-33.
33. Nagappan N, Madhanmohan R, Gopinathan NM, Stephen SR, Pillai DDM, Tirupati N. Oral health-related quality of life and dental caries status in children with orofacial cleft: an Indian outlook. *J Pharm Bioallied Sci* 2019; 11(Suppl 2): S169-S74.
34. Ajami S, Toraby F, Shavakhi M, Eslami N. The impact of type-d personality on oral health-related quality of life in cleft lip and palate adolescents. *J Craniofac Surg* 2018; 29(2): 289-92.
35. Aravena PC, Gonzalez T, Oyarzun T, Coronado C. Oral health-related quality of life in children in chile treated for cleft lip and palate: a case-control approach. *Cleft Palate Craniofac J* 2017; 54(2): e15-e20.
36. Vuletic M, Marcinko D, Vrazic D, Milosevic M, Dediol E, Knezevic P. Development of a valid and reliable instrument for the assessment of quality of life in adolescents with clefts - detection of potential mental health issues. *Psychiatr Danub* 2017; 29(3): 360-8.
37. Broder HL, Wilson-Genderson M, Sischo L. Examination of a theoretical model for oral health-related quality of life among youths with cleft. *Am J Public Health* 2014; 104(5): 865-71.
38. Eslami N, Majidi MR, Aliakbarian M, Hasanzadeh N. Oral health-related quality of life in children with cleft lip and palate. *J Craniofac Surg* 2013; 24(4): e340-3.
39. Ali MA, Abass SK, Nasir EF. A comparative assessment of oral health-related quality of life of children born with orofacial clefts in Sudan and their caregivers'. *BMC Oral Health* 2021; 21(1): 148.
40. Fowler PV, Ayrey SL, Stone PR, Thompson JMD. A nationwide survey of oral health related quality of life of children with orofacial cleft in New Zealand. *Cleft Palate Craniofac J* 2021; 58(8): 1040-6.
41. Nolte FM, Bos A, Prah C. Quality of life among Dutch children with a cleft lip and/or cleft palate: a follow-up study. *Cleft Palate Craniofac J* 2019; 56(8): 1065-71.
42. Broder HL, Wilson-Genderson M, Sischo L. Oral health-related quality of life in youth receiving cleft-related surgery: self-report and proxy ratings. *Qual Life Res* 2017; 26(4): 859-67.
43. Ward JA, Vig KW, Firestone AR, Mercado A, da Fonseca M, Johnston W. Oral health-related quality of life in children with orofacial clefts. *Cleft Palate Craniofac J* 2013; 50(2): 174-81.
44. Pisek A, Pitiphat W, Chowchuen B, Pradubwong S. Oral health status and oral impacts on quality of life in early adolescent cleft patients. *J Med Assoc Thai* 2014; 97 Suppl 10: S7-16.
45. Stock NM, Ridley M. Young person and parent perspectives on the impact of cleft lip and/or palate within an educational setting. *Cleft Palate-Craniofac J* 2018; 55(4): 607-14.
46. Jokovic A, Locker D, Stephens M, Kenny D, Tompson B, Guyatt G. Validity and reliability of a questionnaire for measuring child oral-health-related quality of life. *J Dental Res* 2002; 81(7): 459-63.



47. Broder HL, Wilson-Genderson M. Reliability and convergent and discriminant validity of the Child Oral Health Impact Profile (COHIP Child's version). *CommunDentistry Oral epidemiol* 2007; 35: 20-31.
48. Broder HL, Wilson-Genderson M, Sischo L. Reliability and validity testing for the child oral health impact profile-reduced (COHIP-SF 19). *JPublic Health Dentistry* 2012; 72(4): 302-12.
49. Montazeri A, Goshtasebi A, Vahdaninia M, Gandek B. The Short Form Health Survey (SF-36): translation and validation study of the Iranian version. *QualLife Res* 2005; 14(3): 875-82.
50. Patrick DL, Topolski TD, Edwards TC, Aspinall CL, Kapp-Simon KA, Rumsey NJ, et al. Measuring the quality of life of youth with facial differences. *Cleft palate-craniofacial J* 2007; 44(5): 538-47.
51. Gkantidis N, Papamanou DA, Karamolegkou M, Dorotheou D. Esthetic, functional, and everyday life assessment of individuals with cleft lip and/or palate. *Bio Med ResInt* 2015; 2015.
52. Nigar A, Naqvi I. Body dissatisfaction, perfectionism, and media exposure among adolescents. *Pakistan J Psychol Res* 2019; 57-77.
53. Quittkat HL, Hartmann AS, Dusing R, Buhlmann U, Vocks S. Body dissatisfaction, importance of appearance, and body appreciation in men and women over the lifespan. *Front Psychiatry* 2019; 10: 864.
54. Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and palate: a systematic review. *Eur J Orthod* 2005; 27(3): 274-85.
55. Liu Z, McGrath C, Hagg U. Changes in oral health-related quality of life during fixed orthodontic appliance therapy: an 18-month prospective longitudinal study. *Am J Orthod Dentofacial Orthop* 2011; 139(2): 214-9.
56. Stock NM, Feragen KB. Psychological adjustment to cleft lip and/or palate: A narrative review of the literature. *Psychol Health* 2016; 31(7): 777-813.
57. Tierney S, O'Brien K, Harman NL, Sharma RK, Madden C, Callery P. Otitis media with effusion: experiences of children with cleft palate and their parents. *Cleft Palate Craniofac J* 2015; 52(1): 23-30.
58. Davies JM, Sleeman KE, Leniz J, Wilson R, Higginson IJ, Verne J, et al. Socioeconomic position and use of healthcare in the last year of life: a systematic review and meta-analysis. *PLoS Med* 2019; 16(4): e1002782.
59. Ward E. Cleft lip and palate in India: determining the socioeconomic factors that influence quality of life and treatment received, with a focus in Rural Nainital District, Uttarakhand State 2014.
60. World Health Organization. The World Health Organization quality of life assessment (WHOQOL): position paper from the World Health Organization. *Social science & medicine* 1995; 41(10): 1403-9.