

Critical Review:  
How is Quality of Life Affected in Children with Velo-pharyngeal Insufficiency?

Shannon Serdar  
M.Cl.Sc (SLP) Candidate  
University of Western Ontario: School of Communication Sciences and Disorders

This critical review examines evidence regarding quality of life (QOL) in children with velo-pharyngeal insufficiency (VPI). All of the studies evaluated in this review were between group designs, which used questionnaires to obtain their data and assess outcome measures. Children with craniofacial disorders often experience VPI, and as a result, their QOL is affected in a variety of ways. Although parents perceive a more affected QOL overall when compared to their children with the disorder, the research suggests that both groups view speech limitations, as well as emotional and social difficulties as the primary factors that contribute to reduced QOL in children with VPI. Recommendations for research and clinical practice are provided.

### *Introduction*

The World Health Organization defines “health” as not only a state of being absent from disease, but also a state where an individual experiences enhanced physical, mental, and social well-being, which leads to overall health and translates into quality of life (QOL) (Damiano, Tyler, Romitti, Momany, Jones, Canady, Karnell, & Murray, 2007). Health related QOL is an important measure that provides data related to the outcome of patient health and wellness (Barr, Thibeault, Muntz, & De Serres, 2007). Over the past two decades there has been an increased awareness regarding the relationship between health and QOL. As a result, there has been an effort across all disciplines to improve QOL, rather than quantity of life in all patients experiencing illness and/or a disorder (Barr et al., 2007).

Children with velo-pharyngeal insufficiency (VPI) experience a variety of challenges, including swallowing and communication deficits. These deficits have the potential to impact QOL and communication interactions overall (Rudnick & Sie, 2008). VPI is largely observed in individuals with cleft palate and other craniofacial anomalies that effect functioning of the velo-pharyngeal valve. VPI is defined as the inability to completely close the velo-pharyngeal sphincter (Fisher & Sommerlad, 2011). This is often caused by a congenital structural defect of the velum or the pharyngeal wall, or it can also be caused by mechanical deficits that prevent closure (Trost-Cardamone, 1989). According to Fisher & Sommerlad (2011), adequate velo-pharyngeal closure plays a significant role in the production of most consonant sounds, with the exception of nasal sounds (*m, n, and ng*). The inability to obtain sufficient closure affects children with VPI in many aspects of their lives (Mani,

Carlsson, & Marcusson, 2010). The primary effects of VPI include nasal air escape, hypernasality, and reduced speech volume, which all contribute to impaired speech intelligibility (Rudnick & Sie, 2008). Secondary effects of VPI include nasal regurgitation of food and liquids, compensatory mis-articulations, and facial grimacing (Rudnick & Sie, 2008). Recent findings suggest that VPI also affects confidence, social development, and overall QOL in children with a variety of craniofacial anomalies (Rudnick & Sie, 2008).

Research in the area of QOL, specifically QOL related to children with VPI is limited. Although, there are a number oral health related QOL measures that have been developed to date, these measures are designed to assess QOL in adult populations (Jokovic, Locker, Stephens, Kenny, Tompson & Guyatt, 2002). Over the past decade, researchers and health care professionals have recognized the need for effective and efficient tools to measure QOL, and its impact on health status and treatment in pediatric populations (Barr et al., 2007). As a result, researchers are currently in the process of developing and testing valid and reliable QOL measures for children with VPI (Barr et al., 2007).

QOL is affected in children with VPI on a variety of levels. However, health care professionals treating children with communication disorders tend to focus on the communication impairment, rather than its impact on the general well-being of the individual (Fisher & Sommerlad, 2011). Speech and Language Pathologists (SLPs) have a responsibility to remain cognizant and conscientious of QOL and it’s the impact on therapy, recovery, and general life functioning in all children with craniofacial disorders and VPI.

## *Objectives*

The primary objective of this paper is to critically evaluate the existing literature to have a better understanding of how QOL is affected in children with VPI. The secondary objective is to provide recommendations for continued research and evidence based clinical practice.

## *Methods*

### Search Strategy

Computerized databases, including PubMed, SCOPUS, Pubget, and CINAHL were searched using the following search strategy: (quality of life) AND (velopharyngeal insufficiency) OR (craniofacial disorders) OR (craniofacial anomalies) OR (cleft palate). The search was limited to articles in English. There were no limitations on the dates of the articles. Although reference lists were examined in the articles retrieved, no further articles were found to contribute to the purpose of this review.

### Selection Criteria

Studies selected for inclusion in this critical review were required to investigate how QOL is affected in children with VPI. No limits were placed on the participant demographics. As long as participants were under 18 years of age, they were considered children for the purpose of this review. Furthermore, participants were not limited to children with VPI, but also included their parents and caregivers.

### Data Collection

Results from the literature yielded three articles congruent with the aforementioned selection criteria. The reviewed studies were all between group designs and outcome measures were assessed using questionnaires.

## *Results*

### Study #1

Few studies have been conducted documenting the impact of VPI on QOL in children. This is mainly due to a limited number of valid and reliable instruments available to measure QOL in this particular population (Barr et al., 2007). This study is one of the first to develop an assessment tool that specifically measures QOL in children with VPI. Barr et al. (2007) conducted a between groups design study and used questionnaires to obtain their outcome measures. This type of study design and method of data attainment yields level 4 evidence. The purpose of this study was to determine if QOL is affected in children with VPI. Participants in the sample included 58 children, aged 5-17 years, and their parents. A total of 29 children diagnosed with VPI

participated in the study, while 29 age matched children without VPI were randomly selected to serve as controls. Two questionnaires were administered and completed by the children and their parents: the Pediatric Quality of Life Inventory, version 4.0 (PedsQL) and the Velopharyngeal Insufficiency Quality of Life Inventory (VPIQL). The PedsQL is a standardized assessment tool that measures deficits in physical, emotional, social and school functioning. The VPIQL is a non-standardized assessment tool that was developed to assess child and parent perceptions of functional, emotional, and social aspects of QOL in patients with VPI. Assessment measures are based on 6 subscales: speech limitations, swallowing deficits, situational difficulties, emotional impact, perceptions by others, and activity limitations.

The study used an analysis of covariance (ANCOVA) to determine if there were differences on the domains of the PedsQL and the VPIQL between parents and children with VPI and parents and children without VPI. Means (SDs) were determined for each response in relation to each domain on the questionnaires. Scale values were obtained by calculating the mean of the scale items. Differences between parent and child responses were calculated by subtracting each of the child's score from their parent's scores. The authors found significant differences for emotional functioning ( $p = 0.20$ ) on the PedsQL and swallowing problems ( $p < .001$ ) on the VPIQL. Parents of children with VPI perceived a more negative effect on QOL for emotional problems, and a less affected QOL for swallowing problems when compared to results obtained from their children and the control group. No significant difference was found for speech limitations ( $p = .05$ ) among the VPI children and their parents. However, the positive means indicate that parents perceived a more affected QOL, secondary to speech limitations. Based on the results, Barr et al. (2007) concluded that children with VPI and their parents perceive an overall greater negative effect on QOL in all domains listed in both questionnaires compared to normal controls and their parents.

### Study #2

QOL measures are often administered to patients in order to gain the individual's perspective on their current health status and its impact on QOL. However, in pediatric populations, administering QOL assessment tools can be challenging. As a result, parental perception of child health and wellness are often used to assess the impact of the disorder on their child's QOL (Warschusky, Kay, Buchman, Hallberg, & Berger, 2002). Since research in this area is limited, it not surprising that this is one of the few studies that used parental perspective to measure QOL in children

with craniofacial disorders. Warchausky et al. (2002), conducted a between groups design study and used questionnaires to obtain their outcome measures. This type of study design and method of data attainment yields level 4 evidence. The study identified two primary purposes. The first purpose of the study was to examine and compare health profiles between two groups of children: children with cleft lip and/or palate (CL/P) and children with other craniofacial anomalies. The second purpose of the study was to examine the relationship between parent perceptions of their children's health related QOL, with a specific focus on physical health, psychosocial adjustment, and family functioning. Participants in the study included 54 children and adolescence with craniofacial anomalies, aged 5 to 18 years, and their parents. Of the 54 participants in the study, 50 percent presented with CL/P, 17 percent presented with syndromes, 9 percent presented with synostosis, and the remaining 24 percent presented with other diagnosis. Child participants and their parents were divided in two groups: children with CL/P and children with other craniofacial anomalies. Parents and guardians completed the Child Health Questionnaire version PF28. The Child Health Questionnaire PF28 is a 28 item version of The Child Health Questionnaire, which consists of 11 subscales and three items measuring global health and behavior, as well recent changes in these domains.

The study used a t-test to compare the means of parent's perceptions of general health and QOL between children with CL/P and children with other craniofacial anomalies, and to assess for statistical differences. Since the study was designed to set the foundation for further research, no statistical correlations were developed for multiple testing. The authors found differences in parent's perceptions of general health and QOL between children with isolated CL/P and children with craniofacial anomalies [ $t(45) = 2.57, p < 0.5, df(45)$ ]. Parental perceptions of health and health related QOL for parents of the CL/P group fell within normal limits. However, parents of the children in the group consisting of diverse craniofacial anomalies perceived greater health concerns overall. In addition to the normative data collected, the authors found a positive correlation between perceptions of physical health and psychosocial adjustment in the group of children with diverse craniofacial anomalies. Parents of these children perceived the greatest health concern and impact on QOL to be poor adjustment, specifically poor psychosocial adjustment in relation to the disorder. Furthermore, parents perceived a positive association between family functioning, (specifically family activities and cohesion), and their child's behavior and mental health in response to the disorder. No significant correlations were found between age or

sex of the participants. Based on the results, Warchausky et al. (2002) concluded that overall parents perceive a negative impact on their child's QOL. However, parents of children with a variety of craniofacial anomalies perceive a greater impact on their child's QOL compared to parents of children with isolated CL/P. Furthermore, the authors suggest further research is warranted and additional measures of health and psychosocial status should be implemented in future studies.

### Study #3

Children with craniofacial disorders are affected by their condition. As a result, they are at an increased risk of developing deficits in physical and mental functioning as well as general well-being, which have the potential to significantly affect overall QOL (Locker, Jokovic, & Thompson, 2005). According to Locker et al. (2005), when assessing the outcomes of a disease or disorder, a health related QOL perspective offers an advantage because it is multidimensional and considers the patient's symptoms, physical functioning, and emotional and social well-being. Locker et al. (2005) conducted a between groups design study and used questionnaires to obtain their outcome measures. This type of study design and method of data attainment yields level 4 evidence. The purpose of this study was to assess how health related QOL is affected in children aged 11-14 years with orofacial disorders. Participants in the study included 39 children (25 boys and 14 girls) with orofacial and craniofacial disorders and 32 children (16 boys and 16 girls) with dental conditions. Of the participants in the orofacial and craniofacial group, 11 had isolated cleft lip or palate, 14 unilateral clefts, 5 bilateral clefts, and 9 other craniofacial anomalies. The participants in the second group were mainly comprised of children with significant dental caries and severe malocclusions. The children in the study completed the Child's Perception Questionnaire (CPQ), which consists of 37 items that encompass four domains: symptoms, functional limitations, emotional well-being, and social well-being.

The study used a median split chi square test to determine the mean and median CPQ scores for the overall sample. The mean and the medium CPQ scores were 27.8 and 23.0. These values were quite small when compared to the possible maximum score of 148. Scores at or above the overall median value of 23.0 were found in 59.0% of the participants in the orofacial group, and 37.5% of participants in the dental group ( $p = .059$ ; median split chi square test). The researchers used a Mann-Whitney  $U$  test to assess and compare results between the two groups regarding individual response items on each domain in the questionnaire.

When individual response items were assessed, the orofacial group had significantly higher scores than the dental group for only five questions: two from the functional limitations domain and three from the social well-being domain. These included: breathing through the mouth ( $p < .01$ ), difficulties with speech ( $p < .01$ ), missing school ( $p < .01$ ), being teased by other children ( $p < .01$ ), and being asked questions about their condition by other children ( $p < .001$ ). When the researchers assessed responses and values from the emotional and social well-being domain, they found 50.7% of children in the dental group reported no emotional concerns, compared to only 41.1% of children in the orofacial group (ns; chi-square test). Furthermore, 59.4% of the dental group reported no problems with social well-being, compared to only 23.1% of the orofacial group ( $p < .01$ : chi-square test). Lastly, the researchers found no difference between the two groups in their ratings regarding the extent to which their condition had affected their overall QOL. In the orofacial group, only seven participants reported that it had some effect on their overall QOL, and one participant reported that his/her QOL was affected significantly, while in the dental group the number of participants providing similar responses was eight and zero (ns: chi-square = 2.65;  $df = 1$ ). Overall, the majority of the participants in both groups reported that their condition had a limited affect on their QOL in general. Based on the results, Locker et al. (2005) concluded that although the orofacial group rated some of aspects of their life lower in comparison to the control group, no difference was noted in overall QOL. However, the study found that appearance and speech continue to play a significant role in reduced QOL in children with craniofacial anomalies. Further research is warranted in order to confirm the results.

### *Discussion*

A review of the studies suggests that generally, children with VPI experience a reduced QOL overall in comparison to children without the disorder. However, limitations in the study design as well as weaknesses in the research methodology create limitations. These limitations often result in reduced strength of the evidence, thus, impacting the legitimacy of the claims presented by the researchers.

#### Levels of Study:

As previously stated, the reviewed studies use a between groups study design and outcome measures were assessed using questionnaires, which yields level 4 evidence. This type of study design and method of data attainment is often criticized because it is non-experimental in nature. Furthermore, it is often considered to yield low quality evidence as well as

invalid and unimportant results (Dollaghan, 2007). However, despite these limitations, researchers suggest that survey research is valuable and plays a significant role in obtaining valid and reliable results regarding the feelings, attitudes, beliefs and characteristics of a specific population (Dollaghan, 2007). As a result, when researching QOL, it is important to recognize that the results obtained in survey research are specific to each participant, and can only be reported rather than observed.

#### Research Methods:

Selection criteria was clearly specified across all three studies for both test and control groups. Although it is difficult to find subjects with VPI, all of the reviewed studies created selection criteria based on age, sex, native language, and characteristics of the disorder. Although the reviewed studies took measures to provide sufficient and effective selection criteria, it was observed that the age range of the participants was either too narrow or too broad. For example, Barr et al. (2007) included participants in their sample ranging in age from 5 to 18 years, while Locker et al. (2005) included participants ranging in age from 11 to 14 years. These (broad and narrow) age ranges have the potential to create inherent inconsistencies in the results since responses provided by younger participants may differ when compared to that of older participants, largely due to differences in comprehension levels. Another significant limitation across all three studies was small sample size. Furthermore, participants in the samples were largely drawn from one location due to convenience (e.g. treatment facilities), making it difficult to generalize the results. Although it is challenging to find subjects that meet the selection criteria of these particular studies, further work needs to be undertaken to ensure a larger sample size and to prevent sampling bias (Locker et al., 2005).

In the studies conducted by Barr et al. (2007) and Warschausky et al. (2002), researchers administered questionnaires to both child subjects and their parents. However, it was observed that the researchers tended to rely on parental report. For example, there were some questions on the questionnaire that children were unable or unwilling to answer. Therefore, the parent's perception was considered of greater value when assessing and obtaining answers for those particular questions. Thus, findings from the studies were more susceptible to potential report biases. In addition, preliminary questionnaires were written using terminology that was thought to be appropriate for children of all ages (Barr et al., 2007). However, researchers noted that younger children had difficulty understanding and/ or maintaining attention throughout the questionnaire. As a result, researchers permitted and

advised parents to assist their children if they had difficulty completing the questionnaire (Barr et al., 2007). Since the researchers did not control for potential parental report biases, there is a strong possibility that report biases could impact child responses, and result in a lesser difference observed in their mean scores (Barr et al., 2007). In contrast, Locker et al. (2005) took precautionary measures to control for potential report bias by consulting with child and adolescent professionals to ensure that the wording of all questions listed in the questionnaire were appropriate and comprehensive.

All three of the critical review studies used standardized questionnaires to obtain their data, which ensures sensitivity, internal consistency, validity, and reliability of the results (Locker et al., 2005). However, Barr et al. (2007) administered two questionnaires to both parents and children within the experimental and control groups. One of the questionnaires: The Pediatric Quality of Life Inventory, version 4.0 (Peds QOL) is a standardized test, while the other questionnaire: the Velopharyngeal Insufficiency Quality of Life (VPIQL) is a non-standardized instrument that is specifically designed for children with VPI (Barr et al., 2007). Although the study used at least one standardized questionnaire, the validity and reliability of the results obtained from the non-standardized instrument are questionable.

### *Recommendations*

Based on the limitations of the current research discussed above, the following recommendations have been made:

- I. Selection criteria for experimental and control groups should include age ranges that are sufficient, yet inclusive in order to prevent inconsistencies in the results and to control for potential biases.
- II. Larger sample sizes, in which participants are chosen from diverse locations in order to control for sampling biases and to improve the strength of the evidence by generalizing the results.
- III. Control for potential report biases by creating and utilizing questionnaires that are suitable for children of all ages, and do not require parental assistance.
- IV. Further research should consider using standardized tests only, which specifically measure QOL in children with VPI in order to improve validity and reliability of the results.
- V. Additional research should include the development of standardized assessment tools

that specifically measure QOL in this particular population.

- VI. In the future, longitudinal studies should be considered in order to assess the full effects of VPI on QOL across the lifespan and at each stage of development.

### *Clinical Implications*

Due to the limitations of the reviewed studies, clinicians should remain cautious when implementing these particular findings into clinical practice. Clinicians who choose to work with pediatric and adolescent populations with VPI should consider the following:

- I. Each patient is unique and has different life experiences, which can affect QOL in a number of ways.
- II. QOL is difficult to measure and it can change at any stage of development in a child's life.
- III. Although standardized questionnaires are limited and may not be easily accessible, clinicians and researchers should consider implementing such tools into their assessment protocols when working with children with VPI.
- IV. SLPs and other medical professionals treating children with VPI must acknowledge and remain cautiousness of the potential implications of QOL on recovery, treatment, and overall functioning of all children with the disorder.

### *Conclusion*

Although the studies provide compelling evidence that suggests that there is a correlation between VPI and QOL in pediatric and adolescent populations, the findings should be interpreted with caution. Furthermore, the limitations of the studies discussed above should be considered before the results are implemented into clinical practice. As a result, more research is needed to clarify and confirm methods of data attainment, as well as validity and reliability of the results.

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