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Oral Health-Related Quality of Life in Children with Orofacial Clefts Who Are Treated at a University Hospital

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ORAL HEALTH-RELATED QUALITY OF LIFE IN CHILDREN WITH OROFACIAL CLEFTS WHO ARE TREATED AT A UNIVERSITY HOSPITAL

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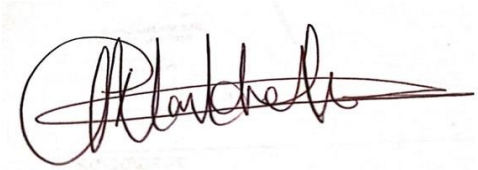
A research report submitted in partial fulfilment of the requirements for the degree of Master of Science in Dentistry, Department of Orthodontics, School of Oral Health Sciences, Faculty of Health Sciences, University of the Witwatersrand, Johannesburg

October 2024

Declaration

I, Tshepang Makhetha, declare that the study entitled “Oral Health-Related Quality of Life in Children with Orofacial Clefts Who Are Treated at A University Hospital” is my own unaided work. It is being submitted for the Degree of Master of Dentistry in the branch of Orthodontics at the University of the Witwatersrand, Johannesburg. It has not been submitted before for any degree or examination at any other University.



… …03 October 2024

Dedication

To my dearest family,

This thesis is dedicated to you, the unwavering source of my strength and inspiration. From a young age, you instilled in me a profound appreciation for knowledge and the transformative power of education. You nurtured my curiosity, encouraged my questioning mind, and provided the unwavering support that allowed me to pursue my academic ambitions.

You demonstrated the true value of learning through your own sacrifices and unwavering commitment to education. You taught me that knowledge is not merely an accumulation of facts but a key to unlocking new worlds, understanding complex ideas, and contributing meaningfully to society. Your encouragement, moments of celebration, and unwavering belief in my potential fueled my determination and guided me throughout this challenging journey.

This thesis stands as a testament to the profound impact you have had on my life. It is a culmination of the countless hours you spent reading to me, answering my endless questions, and fostering my love for learning. I am forever grateful for your unwavering belief in me, even when I doubted myself.

In addition, I would like to dedicate this work to the memory of my late grandmother, Nomthetho Virginia Lydia Modikoane, a nurse. Her strength, resilience, and unwavering love continue to inspire and guide me in all aspects of my life. I am grateful for my time with her and the lessons she taught me. I know she would be proud of the work I have accomplished. With every page of this thesis, I strive to honour your legacy and the values you instilled. I hope this work will inspire others, demonstrating the transformative power of education and the unwavering love and support of a family.

Abstract

Background: Orofacial cleft is a congenital malformation of the palate, lip, or both lip and palate that affects the quality of life in children.

Objectives: To evaluate the oral health-related quality of life (OHRQoL) of children with orofacial clefts treated at a university hospital.

Materials and Methods: A qualitative study using a structured oral health-related quality of life (OHRQoL) questionnaire was conducted. The subscale scores were summarised to obtain the overall OHRQoL score. Concordance between parents and children was examined using a paired sample t-test by comparing the overall and subscale scores. Pearson's correlation coefficients and complementary regression analysis also examined the predictive value of OHRQoL for general health. Cronbach's alpha was used to determine internal consistency, and the subscale score data were statistically analysed. All the tests were at a 5% level of significance.

Results: A total of 39 patients with orofacial clefts participated in this study. Most of the participants were male (22/39; 56%). Most participants were younger than 18 years (28/39;71.8%), presented with cleft lip and palate (24/39; 61.5%) and have not received orthodontic braces (30/39;76.9%).

Cronbach's alpha reliability for each subscale ranged from 0.661 to 0.908, with an overall reliability of 0.898, showing that the instrument was consistent in its measurement.

On average, the overall OHRQoL score was 198.12 (standard deviation: ±0.57), ranging from 38 to 228. The presence of braces had a significant correlation ($p = 0.029$) with general health, and the presence of a cleft had a significant correlation ($p = 0.026$) with oral problems. Participants' ages were shown to significantly correlate with emotion ($p = 0.003$), schooling ($p = 0.012$), the impact on their families ($p = 0.037$), and general health ($p = 0.022$). Most participants had a cleft lip with or without a palate (61.5%), followed by a cleft lip (23.1%), and a cleft palate (15.4%). Cleft lip prevalence was higher in males (77.8%) than in females (22.2%). Overall, 79.5% of adults in the study reported good or great health. A smaller percentage of adults reported average health (10.3%) or were unsure of their health status (10.3%).

Conclusion: Cleft lip and palate were reported in 39 patients, while 30 of them did not wear braces due to which their OHRQoL was not satisfactory. Some of the underlying causes are

the severity of the cleft condition, lack of access to proper and advanced medical services, and socioeconomic factors. These patients reported attributes such as severe pain, eating and speaking difficulties, embarrassment and social isolation that affected their everyday lives. The patients as well as the caregivers recorded high levels of stress and anxiety emphasizing the need to embrace orthodontic treatment and the need to address the general well-being of the patients through socio-psychosocial support to enhance their OHRQoL.

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To my esteemed supervisor, Professor. P. Hlongwa I would like to express my heartfelt gratitude for your invaluable guidance, support, and patience throughout the process of writing my research. Your contributions have been instrumental in shaping this project and ultimately bringing it to fruition.

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I am deeply indebted to you both for your unwavering dedication to my success. This thesis would not have been possible without your invaluable contributions. I am incredibly fortunate to have had you as my supervisors, and I will forever cherish the knowledge and experience I gained from working with you.

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Table of Contents

Declaration .....

Dedication ii

Abstract iii

Acknowledgements v

Table of Contents vi

List of Figures viii

List of Tables ix

List of Abbreviations and Acronyms x

Introduction and Literature Review 1

Orofacial clefts 1

Classification of orofacial cleft 1

Prevalence of orofacial clefts 2

Aetiology of orofacial clefts 2

Diagnosis of cleft lip and palate 3

Orofacial clefts effect on patients 4

Management of orofacial clefts 5

Health-related quality of life in orofacial clefts 6

Measurement of health-related quality of life in orofacial clefts 7

Statement of purpose 10

The aim of the study 10

The objectives of the study 10

Materials and Methods 11

Study design 11

Study settings 11

Sample size 11

Data collection 11

Data analysis 12

Ethical considerations 12

Results **14**
Demographic information of the sample 14
Correlation analysis of OHRQoL subscales 18
Oral symptoms and functional wellbeing. 19
Emotional wellbeing 20
Schooling 21
Peer relationship 22
Family impact 23
General health 24
Discussion **26**
Conclusions and Recommendations **32**
Conclusions 32
Recommendations 33
References **34**
Appendices **42**
Appendix 1: Oral Health-Related Quality of Life Questionnaire **42**
Appendix 2: Ethical Clearance Certificate **46**
Appendix 3: Permission Letter from the CEO at WOHC **47**
Appendix 4: Participants' Information Sheet: Parents/caregivers **48**
Appendix 5: Participants' Information Sheet: Children Under 18 Years of Age **51**
Appendix 6: Consent Form: Adult participants/parents/caregivers **54**
Appendix 7: Children Assent Form **56**
Appendix 8: OHRQoL scores **58**
Appendix 9: Turnit-in Report **61**

List of Figures

Figure 1: Relationship network for participants with cleft lip and palate 16

Figure 2: General health of participants based on the cleft type 17

Figure 3: Distribution of general health among participants with OFC 25

List of Tables

Table 1: Overview of study objectives and statistical analysis 12

Table 2: Sociodemographic and oral health status distribution of patient 14

Table 3: Cronbach's alpha scores for each section of the questionnaire 15

Table 4: Mean overall and sections score for participants. 17

Table 5: Spearman rho correlation coefficients between the OHRQoL sections 19

Table 6: Item descriptive statistics for oral symptoms and functional wellbeing 20

Table 7: Item descriptive statistics for emotional wellbeing 21

Table 8: Item descriptive statistics for schooling 22

Table 9: Item descriptive statistics for peer relationship 23

Table 10: Item descriptive statistics for family impact 24

List of Abbreviations and Acronyms

CEO	Chief Executive Officer
CL/P	Cleft Lip/Palate
DNA	Deoxyribonucleic Acid
HRQoL	Health-related Quality of Life
IRF6	Interferon Regulatory Factor 6
LAHSHAL	Lip (L), alveolus (A), hard palate (H), soft palate (S), H (Hard Palate Sn), A (SN Alveolus), L (Sn Lip).
MID1	Midline 1
MSX1	Msh Homeobox 1
OFC	Orofacial Cleft
OHRQoL	Oral Health-Related Quality of Life
PHF8	Plant Homeodomain Finger Protein 8
PVRL1	Poliovirus Receptor-related 1
SD	Standard Deviation
TBX22	T-box Transcription Factor-22
TGFB3	Transforming Growth Factor-Beta-3
WHO	World Health Organization
WOHC	Wits Oral Health Centre

Introduction and Literature Review

Orofacial clefts

Orofacial clefts (OFCs) are congenital malformations of the palate, lip, or both lip and palate (Nasreddine et al., 2021). OFCs may be an opening in the lip, known as a cleft lip; it may also involve the roof of the mouth, known as the hard palate, or the soft tissue in the back of the mouth, known as the soft palate, which is known as a cleft palate; or it may involve the lip and the palate, known as a cleft lip and palate (CL/P;(Lewis et al., 2017). The opening on the lip may be on one side (unilateral), both sides (bilateral), or in the middle of the lip (midline cleft). In other cases, OFCs may involve structures around the oral cavity that can extend to other facial structures, leading to oral, facial, and craniofacial deformity (Mossey and Little, 2009).

Classification of orofacial cleft

The classification system of OFCs varies depending on the presence of other anomalies, familial or non-familial inheritance, and genetic or environmental factors. The classifications can either be postnatal (anatomical location and extent of the cleft) or sonographic clefts (classify clefts based on their location and extent). The postnatal Spina-A classification system uses the incisive foramen as an anatomical reference to divide malformations into four groups (I–IV) (Rodrigues et al., 2018). The postnatal LAHSHAL (L (Right Lip); A (Right Alveolus); H (Right Hard Palate); S (Soft Palate); H (Hard Palate Sn); A (SN Alveolus); L (Sn Lip) classification system uses symbols and letters to record the degree of completeness and laterality of clefts. Postnatal classification divides clefts into subgroups based on their morphology (Nasreddine et al., 2021).

A prenatal examination of the foetal face and brain on an ultrasound can help increase detection rates of OFCs (Salomon et al., 2011, Salomon et al., 2013). Antenatal diagnosis of cleft palate is difficult, especially in the first trimester, and thus, requires antenatal sonographic classification (Mak and Leung, 2019). Antenatal sonographic classification can identify different cleft types, such as type 1 isolated cleft lip alone, type 2 unilateral CL/P, type 3 bilateral CL/P, type 4 median CL/P, and type 5 clefts associated with amniotic bands or limb-body-wall-complex, which have fatal outcomes (Smarius et al., 2017).

Prevalence of orofacial clefts

OFCs are estimated to occur in 1 in 500–700 births worldwide, with a global prevalence of about 9.92 per 10 000 live births (Massenburg et al., 2016, World Health Organization, 2016, Kantar et al., 2019). The prevalence of OFCs varies by region and ethnicity, with Asians having the highest prevalence (1 in 500), followed by Caucasians (1 in 1 000) and Africans (1 in 2 500; (Mossey and Little, 2009, Mukhopadhyay et al., 2021, Nasreddine et al., 2021). In Africa, the prevalence of OFCs ranges from 0.3 to 1.65 per 1 000 live births, with a prevalence rate of 0.33 per 1 000 live births in South Africa (Panamonta et al., 2015). It is important to note that the prevalence of OFCs may be underreported in some low-income countries due to limited access to healthcare services and birth registration.

OFCs are more common in males than in females. In addition, males are more likely to have a cleft lip or CL/P, and females present more with cleft palate (Hlongwa et al., 2019).

Aetiology of orofacial clefts

The aetiology of OFCs is polygenic and multifactorial, involving various genetic and environmental factors (Murray and Wehby, 2010, Muhamad et al., 2014). Orofacial clefts can occur alone (non-syndromic) or in combination with other birth defects (syndromic). Non-syndromic OFCs are the most common, accounting for about 70% of cases. Syndromic OFCs are associated with several different genetic syndromes, each with its unique combination of birth defects (Setó-Salvia and Stanier, 2014). Research into CL/P syndromes shows that the disease has a significant genetic basis, as evidenced by strong familial aggregation. This means that individuals with a family history of CL/P are at an increased risk of being born with the condition (Beaty et al., 2016). Abnormal gene variants inherited from the mother or father may be directly responsible for the cause of the CL/P or confer susceptibility to developing a cleft (Setó-Salvia and Stanier, 2014).

Some specific genes that have been identified as causing CL/P include T-box transcription factor-22 (TBX22), poliovirus receptor-like-1 (PVRL1), and interferon regulatory factor-6 (IRF6; (Kohli and Kohli, 2012, Askarian et al., 2022). X-linked genes such as midline 1 (MID1) on chr Xp22, TBX22 on chr Xq21.1, and PHD finger protein 8 (PHF8) on chr Xp11.22 are strongly associated with syndromic clefts (Jugessur et al., 2012). Whereas the transforming growth factor-alpha (TGFA), transforming growth factor-beta-3 (TGFB3), 5,10 methylenetetrahydrofolate reductase (MTHFR), and msx homeobox homolog-1 (MSX1) have

been linked to non-syndromic cleft lip and palate (Kohli and Kohli, 2012). Alterations in one or more of these genes (e.g., IRF6, MSX1, PVRL1) could result in cleft lip, cleft palate, or CL/P (Saleem et al., 2019).

A study showed that facial tissues are derived from the neural crest, and therefore, periconceptional folic acid supplementation may reduce the occurrence of OFCs in offspring (Agarwal and Gopalan, 2011). Studies identified the potential associations between a high incidence of OFCs and inadequate family planning, a low-folate diet, and inadequate folate use during the periconceptional period and the first trimester of pregnancy (Wehby et al., 2012a, Figueiredo et al., 2015, Gatt et al., 2016). In addition to folic acid, other nutrients and vitamins such as zinc, riboflavin, and vitamin A are also essential for foetal development and may play a role in preventing OFCs (Agarwal and Gopalan, 2011). A deficiency of folic acid, zinc, riboflavin, vitamin A, copper, or manganese during pregnancy has been reported to be the cause of cleft palate and other malformations in animal studies (Maldonado et al., 2021).

Maternal age, certain medications, smoking, and alcohol consumption during pregnancy are all risk factors for OFCs (Campos Neves et al., 2016). Maternal age is a risk factor for OFCs, with the risk increasing slightly after age 35. This is thought to be due to a decline in egg quality and an increase in chromosomal abnormalities with age. Medications such as anti-epileptic drugs and corticosteroids can increase the risk of OFCs, especially if taken during the first trimester of pregnancy. Corticosteroids used to treat asthma, inflammation, and other conditions can also increase the risk of OFCs, but the risk is lower than with anti-epileptic drugs (Xiao et al., 2017, Peterka et al., 2021). Smoking and alcohol consumption during pregnancy are also risk factors for OFCs because smoking can reduce blood flow to the foetus and damage DNA, and alcohol can interfere with the development of the foetus's face (Garland et al., 2021).

Diagnosis of cleft lip and palate

A cleft lip can be detected on ultrasound as early as 13 weeks of gestation. A cleft palate is more difficult to see on ultrasound, but it can be detected in about half of cases by 22 weeks of gestation. If an ultrasound shows a possible cleft, the doctor may recommend further testing, such as amniocentesis and/or chorionic villus sampling (Nicolaides, 2003). The accuracy of transabdominal ultrasound in detecting cleft lip and palate at 16 weeks gestation has not been well-established, even though the foetal face can be successfully imaged at this time (Shaikh

et al., 2001). Minor clefts, such as submucous cleft palate and bifid uvula, are sometimes diagnosed later in life because they may be difficult to detect at birth (Hanny et al., 2016).

The submucous cleft palate is a type of cleft palate that occurs in the soft palate, which is the back part of the roof of the mouth. It is often covered by the lining of the mouth, making it difficult to see. Bifid uvula is a condition in which the uvula, a small flap of tissue at the back of the throat, is split. It is also often difficult to see at birth. A cleft lip is easily diagnosed with ultrasonography in the second trimester of pregnancy if the position of the foetus's face is located correctly (Murray et al., 1997). The prenatal diagnosis gives parents the advantage of having time to prepare emotionally for the birth and become knowledgeable about the birth defect (Crockett and Goudy, 2014). Cleft lip and cleft palate are usually diagnosed at birth because doctors examine babies' faces and mouths for any signs of a cleft.

Orofacial clefts effect on patients

OFCs may negatively impact individuals' health as they are associated with frequent ear infections and may also result in speech difficulties due to nasal escape articulation problems (Agbenorku, 2013). Hearing difficulties due to abnormalities in the palatal musculature; feeding difficulties at birth due to problems with an oral seal, swallowing, and nasal regurgitation; and dental problems have been reported (Agbenorku, 2013). Individuals with OFCs are at increased risk of developing communication delays and disorders, which can impact their interactions with others, including their perceptions of themselves and their relationships with others.

Children and adolescents with OFCs have an increased risk of developing psychiatric disorders, as noted by high levels of depression (Demir et al., 2011). Children and adolescents with CL/P are also found to show sleep disturbances more often than those without CL/P (Ho et al., 2023). Studies found that children and adolescents born with OFCs face negative effects, including being stared at, talked about by others, and teased about their appearance (Kapp-Simon and Gaither, 2016, Breseke, 2017).

Many parents of children with OFCs commonly express a loss of control over their plans for a healthy pregnancy. Mothers often report resentment, hurt, and disappointment after discovering their child has an OFC (Hlongwa and Rispel, 2018). The psychosocial wellbeing of parents is also impacted by the psychological problems their children with OFCs may develop. Children with OFCs are at greater risk of anxiety disorders, hyperactivity disorders,

and academic achievement problems than unaffected children (Wehby et al., 2012b, Tyler et al., 2013).

Management of orofacial clefts

Managing a patient with an OFC is very complex as it requires a multidisciplinary team, including inter alia orthodontists, maxillofacial surgeons, otolaryngologists, plastic surgeons, paediatricians, and speech-language pathologists. Every child with a OFC should also be evaluated by a medical geneticist at some point to determine whether the defect is an isolated occurrence or if it is part of a genetic syndrome (Robin et al., 2006). The surgical repair of the cleft palate is often performed after the age of 9–10 months as this is associated with optimal speech outcomes (Mahoney et al., 2013). Furthermore, the techniques used for cleft palate repair vary depending on the length and width of the cleft as well as the type of cleft palate according to various classifications (MacKay, 2012).

To achieve the best results, surgical treatments should be performed within three months for cleft lip repair and between 6 and 12 months for cleft palate surgery (Ziak et al., 2010, Raghavan et al., 2018). Many surgeons favour surgery for unilateral cleft lips in children weighing up to 4.5 kg with hemoglobin levels under 8 gm%, while bilateral clefts often require a weight of 5-6 kg and hemoglobin levels below 9 gm% (Pendem et al., 2024). Cheiloplasty, palatoplasty, and alveolar bone grafting are the three basic surgeries scheduled for the treatment of a cleft deformity in the common cleft lip and palate patient with cleft alveolus (Vandersluis et al., 2020, Dunworth et al., 2022). Lip revision, velopharyngeal dysfunction correction, orthognathic surgery for maxillary hypoplasia, rhinoplasty for nasal deformity, and velopharyngeal incompetence surgery are some of the secondary treatments that can be performed later for patients (Scopelliti et al., 2013, Pai et al., 2019).

Hearing, speech and language development in patients with OFCs must be thoroughly screened both before and after surgery, taking into account all physical, psychological, and linguistic factors associated with the structural anomaly (Russell, 2010). Speech therapy or surgery is necessary for 20% of children to achieve intelligible speech, regular middle ear disease check-ups are required to tackle otologic problems, and orthodontic treatment is required to treat maxillary collapse (Lane et al., 2022).

Psychological support for individuals affected (patients and parents) by OFCs should commence promptly following diagnosis, including prenatal diagnoses (Kramer et al., 2007).

This benefits both the patient and their family by helping them visualise and plan for the baby, discuss feeding options (especially breastfeeding), learn about the timing and type of surgery required, and plan (Binford et al., 2022). Since OFC treatment involves both surgical and non-surgical procedures from childhood until adulthood, it can have a significant psychological impact on both the patient and their family members (Turner et al., 1997). Individuals with OFCs often experience low self-esteem and social skills challenges, especially girls (Branson et al., 2024). A child's speech can have a profound impact on their social interactions and overall well-being (Tannure et al., 2013). Parents need reassurance, support, and time to process the information so they can provide their child with the best possible support and care. Psychological care can help parents develop strong support networks and prevent the development of a negative self-concept in their child. It is also important for parents to discuss with their children ways to handle negative social situations related to their OFC (Berger and Dalton, 2011).

The OFC defects come with a high burden of diseases because of the complexities and effects arising with all aspects of the lives of both the patients and their families (Zeraatkar et al., 2019). Furthermore, the success of medical treatment of children with a cleft is not only measured by mending the cleft but also extends to the maintenance and improvement of the child's quality of life after surgery (Opriş et al., 2022). Children born with OFCs can present with craniofacial abnormalities and chronic health conditions that require a prolonged treatment protocol, and therefore, evaluating their quality of life has become an important part of evaluating health programs.

Health-related quality of life in orofacial clefts

OFCs are referred to as an impairment since there is a structural or functional loss in the body as described by the *International Classification of Functioning and Disability 2* developed by the World Health Organization (WHO) in 1990 (Neumann and Romonath, 2012). Patel and Ross (2003) suggest that it is important to study the quality of life in individuals with OFCs from the theoretical perspective of the *International Classification of Functioning and Disability 2* framework as it provides insight into understanding how the impairment affects individuals' activities and participation in society.

The World Health Organization Quality-of-Life Scale (WHOQOL) Group (1994) describes the quality of life of an individual as their physical and mental health, their interpersonal relationships with others, their environment, and their ability to function independently. The concept of health-related quality of life (HRQoL) is defined as "the individual perspective of the impact of their health status including, disease and treatment on psychological, psychical and social functioning" and is increasingly received attention in health care (Kitson et al., 2012). Interventions that incorporate patient-reported outcome measurements to evaluate patients' Oral Health Related-Quality Of Life (OHRQoL) in research, clinical practice, and service evaluations can target aspects of care that are most important to the patient (Kitson et al., 2012). Patient-reported outcome measurements can either be condition-specific or generic, with the latter evaluating the aspects relevant to the medical condition (Lang et al., 2012).

Previous studies report decreased quality of life in adolescents with congenital and acquired facial malformations compared with unaffected adolescents (Masnari et al., 2012, Meyer, 2021). It was shown that CL/P can adversely affect emotional and social functioning, increase the financial burden on the family, affect the family income, lower marriage rates, increase social anxiety due to speech problems, and increase mortality rates due to suicide (Wehby and Cassell, 2010).

Measurement of health-related quality of life in orofacial clefts

Oral health-related quality of life (OHRQoL) before and after treatment can be assessed by the following tools: WHO Quality of Life assessment instrument, Pediatric Quality of Life Inventory, 36-Item Short Form Health Survey Questionnaire, Oral Health Impact Profile, Child Oral Health Impact Profile, Michigan Oral-Health Related Quality of Life Scale, Child Perceptions Questionnaire, Children's Health and Illness Profile Adolescent Edition, and Child Health Questionnaire Parent Form 28 (De Queiroz Herkrath et al., 2015). Several studies investigated the quality of life for adults and adolescents with oral clefts (Marcusson et al., 2001, Strauss and Fenson, 2005, Topolski et al., 2005). The current study is important because it will fill the gap in research on the OHRQoL of children with oral clefts in South Africa to better understand the impact of oral clefts on their and their families' lives, identify their specific needs, and develop and evaluate tailored interventions and support services.

The WHO Quality of Life is a generic quality-of-life measure that was developed by the WHO in the early 1990s. It is a 100-item questionnaire that assesses four domains of quality of life,

namely physical health, psychological health, social relationships, and environment (WHO, 1998). To address the need for a faster tool for large-scale epidemiological studies, the WHO created a shorter 26-question version of its quality-of-life measure, called the WHO Quality of Life-BREF. The WHO Quality of Life has been translated into over 100 languages and is used in research and clinical practice around the world.

The Pediatric Quality of Life Inventory is a generic quality-of-life measure that was developed specifically for children and adolescents (Varni et al., 1999). It is a 23-item questionnaire that assesses four domains of quality of life, namely physical functioning, emotional functioning, social functioning, and school functioning. It allows researchers to compare the OHRQoL of children with different diseases, as well as compare the OHRQoL of children with specific diseases to children who are healthy (Matza et al., 2004). It has been translated into over 30 languages and is used in research and clinical practice around the world.

The 36-Item Short Form Health Survey Questionnaire is a generic quality-of-life measure that was developed by the RAND Corporation in the early 1980s (Ware and Sherbourne, 1992). It is a 36-item questionnaire that assesses eight domains of health, namely physical functioning, role limitations due to physical health problems, bodily pain, general health perceptions, vitality, social functioning, role limitations due to emotional problems, and mental health. It has been translated into over 50 languages and is used in research and clinical practice around the world.

The Oral Health Impact Profile is a condition-specific quality-of-life measure that was developed to assess the impact of oral health problems on people's daily lives. This is the most widely used tool for measuring the negative effects of oral health problems on individuals' overall well-being and quality of life. A 49-question version of the instrument was created and tested, which was later shortened to 14 questions that maintain the same dimensions (Slade, 1997). The seven dimensions of oral health impact assessed are functional limitations, pain, psychological discomfort, physical disability, social disability, handicap, and financial impact. It has been translated into over 30 languages and is used in research and clinical practice around the world.

The Child Oral Health Impact Profile is a condition-specific quality-of-life measure that was developed to assess the impact of oral health problems on the daily lives of children and adolescents (Broder et al., 2012). The purpose of the questionnaire was to evaluate school-age children's oral-facial well-being based on their own reports as well as proxy reports from caregivers. It is a 34-item questionnaire that assesses the impact of five domains of oral health, namely oral health, functional wellbeing, social/emotional wellbeing, school environment, and self-image (Broder et al., 2012). Because of its validity and reliability, which have been demonstrated in cross-cultural adaptations, it has been translated into numerous languages and is utilised in research and clinical practice all over the world (Myint et al., 2020).

The Michigan Oral-Health Related Quality of Life scale is a condition-specific quality-of-life measure that was developed to assess the impact of oral health problems on the daily lives of people of all ages (Broderick et al., 2000). It is a 10-item questionnaire that assesses five domains of oral health impact, namely oral function, pain, emotional wellbeing, social wellbeing, and satisfaction with oral health. It has been translated into over 10 languages and is used in research and clinical practice around the world.

The Child Perceptions Questionnaire is a condition-specific quality-of-life measure that was developed to assess the impact of oral health problems on the daily lives of children (age 8–10) and adolescents (age 11–14). It is a 12-item questionnaire that assesses two domains of oral health impact, namely oral symptoms and impact on daily living. It has been translated into over 20 languages and is used in research and clinical practice around the world.

The Children’s Health and Illness Profile Adolescent Edition is a generic quality-of-life measure that was developed to assess the quality of life of adolescents with chronic illnesses (Williams et al., 1993). It is a 52-item questionnaire that assesses six domains of quality of life, namely physical health, emotional health, social health, self-esteem, family functioning, and school functioning. It has been translated into over 10 languages and is used in research and clinical practice around the world.

The Child Health Questionnaire Parent Form 28 is a generic quality-of-life measure that was developed to assess the quality of life of children and adolescents from the perspective of their parents (Landgraf et al., 1996). It is a 28-item questionnaire that assesses four domains of quality of life, namely physical health, emotional/behavioural health, self-esteem, and family functioning. It has been translated into over 20 languages and is used in research and clinical practice around the world.

Statement of purpose

OFCs are congenital malformations commonly diagnosed at birth and may have a negative impact on the health of an individual, subsequently creating a financial burden on the affected family. The cleft clinic at the Wits Oral Health Centre (WOHC) has been operating for more than nine decades. Children with OFCs at WOHC have not been evaluated on the OHRQoL effects this condition has on them. Therefore, this study evaluated the OHRQoL in children with OFCs treated at WOHC.

The aim of the study

The study had the following aim: To evaluate the impact of OFC on the OHRQoL of children with OFCs treated at WOHC.

The objectives of the study

This study had the following objectives:

- To determine the demographic profile of children with OFCs treated at WOHC;
- To evaluate the OHRQoL of the children with OFCs consulted for orthodontic treatment at WOHC; and
- To determine the factors associated with OHRQoL of children with OFCs.

Materials and Methods

Study design

This was a qualitative study that used a structured OHRQoL questionnaire to collect data.

Study settings

The study was conducted at the cleft clinic of the WOHC. Cleft lip and palate patients consulted for orthodontic treatment were recruited to participate in the study, and when a patient was a minor, the parents/caregivers were asked to complete the questionnaire. For participants who could not write, the principal investigator assisted the patient.

Sample size

A convenient sample size was used with an estimated number of 60 clinical records of CL/P patients that were consulted at WOHC by postgraduate students. This number was based on the number of records available during the study and the number of working postgraduate students which was sufficient to adequately review and consult records at WOHC. All patients were included in the sample as the number was too small for sampling.

Data collection

The OHRQoL questionnaire used in this study was a modified Child Oral Health Impact Profile questionnaire (Appendix 1) adapted from Geels et al. (2008). The questionnaire was written in English; if the patient did not understand English, the questionnaire was verbally clarified in the South African official language the patient understands. The principal investigator (TM) is fluent in all South African languages. The questionnaire took approximately 15 minutes on average to complete per patient.

The questionnaire administered in this study comprised six distinct sections, with identical versions for children and parents. These sections covered a wide range of aspects related to the children's oral health and overall wellbeing. The first section gathered demographic information about the participants. Following that, there were sections dedicated to assessing oral symptoms and functional wellbeing (10 items), emotional wellbeing (10 items), schooling (5 items), peer relationships (5 items), family impact (9 items), and general health (1 item)

(**Appendix 1**). A 6-point Likert scale was used in this study to capture the response to the questionnaire items as follows: 1 = *never*, 2 = *sometimes*, 3 = *regularly*, 4 = *usually*, 5 = *always*, and 6 = *uncertain*. Furthermore, a separate question about the child's general health was added to the questionnaire for parents'/caregivers' responses with the following responses: 1 = *bad*, 2 = *poor*, 3 = *average*, 4 = *good*, 5 = *great*, and 6 = *uncertain*.

Data analysis

For each section (B, C, D, E, F), Cronbach's alpha was used to determine internal consistency, and values of > 0.7 were considered acceptable (Gilchrist et al., 2014). The data were cleaned by handling missing values and *uncertain* responses in the same way. The items were ordered with several *uncertain* responses to make them easier to analyse. Then, the adjustment method was used to replace *uncertain* responses with the personal mean on that subscale. This method was chosen because it performs well compared to other methods of missing data imputation (Shrive et al., 2006; Geels et al., 2008). Subscale scores were determined by summarising the responses of all items of each section. The addition of the subscale scores yielded the overall OHRQoL. Pearson's correlation coefficients and complementary regression analysis also examined the predictive value of OHRQoL for general health. Table 1 gives an overview of the objectives and statistical analysis.

Table 1: Overview of study objectives and statistical analysis

Study objectives	Analysis
1. To determine the demographic profile of children with OFCs treated at WOHC	Descriptive statistics
2. To evaluate the OHRQoL of the children with OFCs consulted for orthodontic treatment at WOHC	Descriptive statistics (calculate mean, median, and mode)
3. To determine the factors associated with OHRQoL of children with OFCs	T-test and Pearson correlation

Ethical considerations

Ethical clearance was obtained from the Human Research Ethics Committee (Medical) of the University of the Witwatersrand, Johannesburg (Ref. M210816; Appendix 2). Permission to access hospital records was received from the CEO of WOHC (Appendix 3).

A participants' information sheet (Appendix 4 & 5) and consent form (Appendix 6) were given to parents/guardians of the children with OFCs when recruiting them to participate in the study. Only children whose parents gave consent were included in the study. The verbal accent was obtained all children participating in the study and recorded in the accent form (Appendix 7).

There was no coercion, and participants could withdraw from the study without prejudice. The principal researcher maintained the anonymity and confidentiality of the captured data by removing personal details and using unique identification codes.

Results

Demographic information of the sample

A total of 39 participants were included in this study. The final sample size was less the predicted convenient sample size of 60 as a result of the COVID-19 pandemic in 2020 as well as the fire disaster that happened at Charlotte Maxeke Johannesburg Academic Hospital in 2021, which affected patients access to WOHC. The participants were composed of more males (22/39; 56.4%) than females (Table 2). Most of the participants (28/39; 71.8%) were younger than 18 years. The clinical presentation of OFC showed a predominance of CL/P (24/39; 61.5%) to cleft lip (9/39; 23.1%) and cleft palate (6/39; 15.4%). During the data collection, more participants did not receive orthodontic braces (30/39; 76.9%) than did receive orthodontic braces (6/39; 23.1%). The patients were predominantly Black (31/39; 79.5%), followed by Coloured (4/39; 10.3%), Indian (3/39; 7.7%), and White (1/39; 2.6%).

Table 2: Sociodemographic and oral health status distribution of patient

Variable	Category	Frequency	Percentage
Gender	Male	22	56.4
	Female	17	43.6
Race	Black	31	79.5
	White	1	2.6
	Coloured	4	10.3
	Indian	3	7.7
Age	< 18	28	71.8
	> 18	11	28.2
Type of cleft	Cleft lip	9	23.1
	Cleft palate	6	15.4
	CL/P	24	61.5
Have braces	Yes	9	23.1
	No	30	76.9

The reliability of the questionnaire as a whole and of each of its sections was measured using Cronbach's alpha. Table 3 shows Cronbach's alpha reliability for each section, ranging from 0.661 to 0.908. The overall reliability of 0.898 indicates that the instrument is reliable, as reliability is generally accepted when Cronbach's alpha is greater than 0.70 (Kilic, 2016). This means that the results of the instrument can be trusted.

Table 3: Cronbach's alpha scores for each section of the questionnaire

Variables	**#Items**	**Cronbach α**
Oral symptoms and functional wellbeing	10	0.661
Emotional wellbeing	10	0.817
Schooling	5	0.749
Peer relationship	5	0.702
Family impact	9	0.908
General health	1	0.900
Completed instrument	40	0.898

The relationship map illustrated in Figure 1 indicates that most participants were Black, did not have braces, and were under the age of 18. The participants also presented predominantly with CL/P.

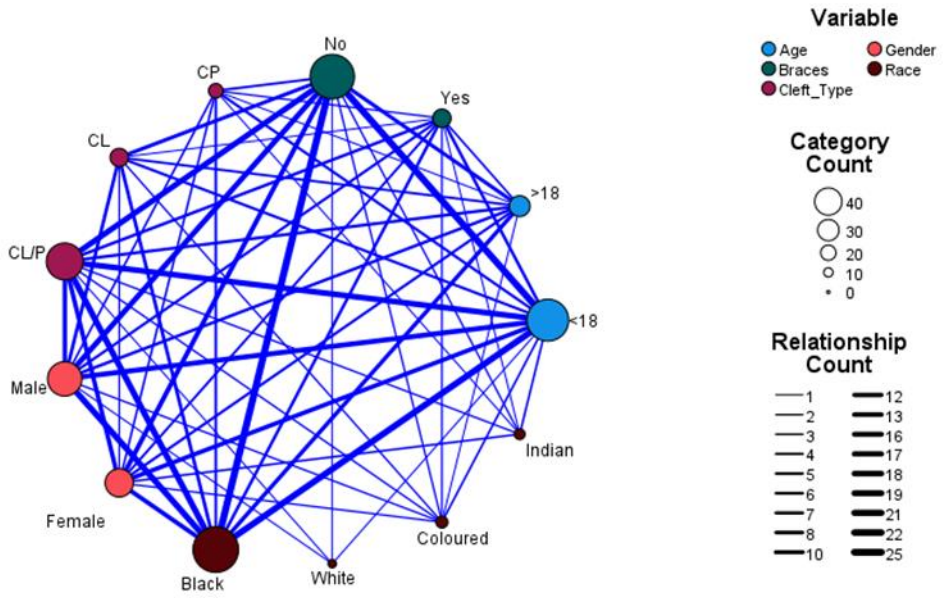

Figure 1: Relationship network for participants with cleft lip and palate

Figure 2 shows the evaluation of the general health of the participants, and each general health factor, such as *average*, *good*, *great*, or *uncertain*, has a combined interpretation of the cleft lip, cleft palate, and CL/P. The bar graph (Figure 2) shows the number of people with *good*, *great*, and *uncertain* general health. The number of people with *good* general health is the highest (18/39; 46.1%), followed by the number of people with *great* general health (13/39; 33.3%), *uncertain* general health (4/39; 10.3%), and then the number of people with *poor* general health (4/39; 10.3%). This suggests that most of the people in the study have *good* general health, while a smaller proportion have *great* or *uncertain* general health.

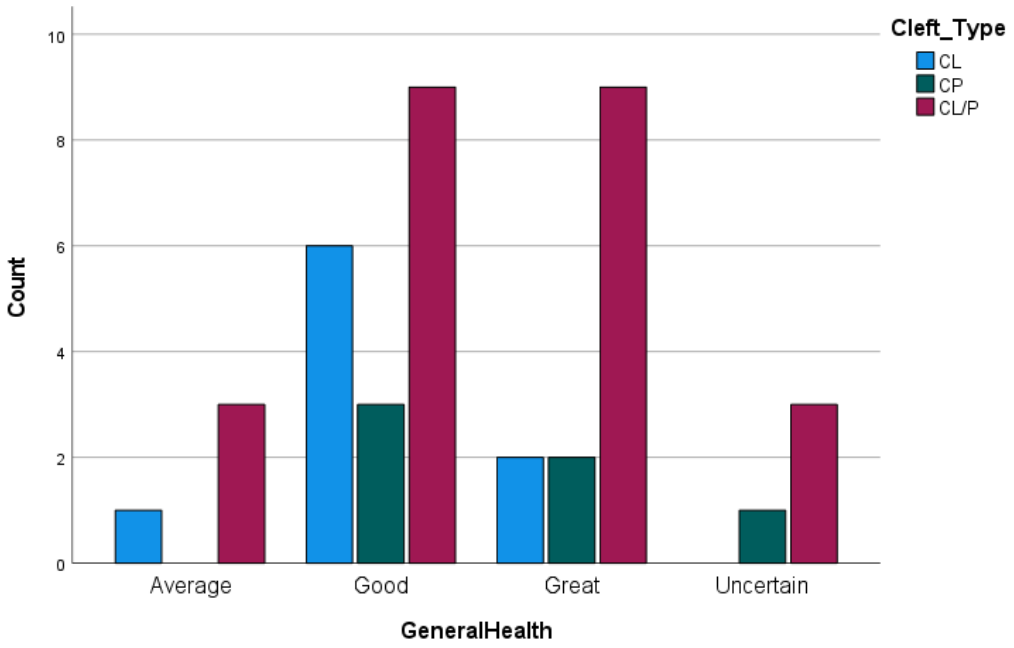

Figure 2: General health of participants based on the cleft type

The mean OHRQoL score for children with OFCs was 198.12 (SD=0.57). The highest score was observed in emotional wellbeing at the mean of 28.27 (SD=0.24), and the lowest was general health with a mean of 4.4 (SD=0.05)

Table 4: Mean overall and sections score for participants.

	N items	**Mean (SD)**
HRQoL (overall) [40–240]	40	198.12 (0.57)
Section		
Oral symptoms and functional wellbeing [10–60]	10	24.56 (0.58)
Emotional wellbeing [10–60]	10	28.27 (0.24)
Schooling [5–30]	5	10.46 (0.38)
Peer relationship [5–30]	5	11.19 (0.44)
Family impact [9–54]	9	24.58 (0.36)
General health [1–6]	1	4.4 (0.05)

Correlation analysis of OHRQoL subscales

The questionnaire was evaluated to assess the correlation between oral health and other aspects of wellbeing, including emotional wellbeing, peer relationships, schooling, and family impact. It also assessed the correlation between oral health and other factors such as gender, age, cleft type, and wearing braces. There was a significant correlation between oral symptoms and functional wellbeing and emotional wellbeing, with a significance of 0.001. The oral symptoms and functional wellbeing also had a significant correlation of 0.003 with peer relationship. Emotional wellbeing had a significant correlation of 0.004 with schooling, 0.001 with peer relationship, and 0.031 with family impact. School was observed to also have a significant correlation with family impact, with a significant correlation of 0.001. General health had a significant correlation of 0.017 with oral symptoms and functional wellbeing. The gender of participants had a significant correlation of 0.035 with general health, while age had a significant correlation with emotion (0.003), school (0.012), family impact (0.037), and general health (0.022). The cleft type had a significant correlation with oral symptoms, with a value of 0.026. The wearing of braces had a significant correlation of 0.029 with general health. The Spearman rho correlation coefficients between the OHRQoL sections are shown in Table 5.

Table 5: Spearman rho correlation coefficients between the OHRQoL sections

	Oral	Emotion	School	Family	Peer	General
Oral	-	0.001**	0.090	0.291	0.003**	0.017
Emotion	0.001	-	0.004**	0.031*	<0.001**	0.285
School	0.090	0.004**	-	0.001**	0.051	0.565
Family	0.291	0.031*	0.001**	-	0.523	0.128
Peer	0.003**	<0.001**	0.051	0.523	-	0.500
General	0.017*	0.285	0.565	0.128	0.500	-

**Correlation is significant at the 0.01 level (2-tailed).
*Correlation is significant at the 0.05 level (2-tailed).

Oral symptoms and functional wellbeing.

Table 6 shows the descriptive statistics (mean, standard deviation, minimum and maximum) of 10 oral symptoms and functional wellbeing items. The mean score for all items is between 1.77 and 3.85, indicating that the participants reported moderate to severe levels of oral symptoms and functional wellbeing problems. The evaluation of oral symptoms and functional wellbeing indicated that 38.5% of the participants never 'had pain in your teeth/toothache', followed by 30.8% responding sometimes. Concerning breathing through the mouth, 20.51% responded to always having the experience and 51.3% with never having the experience. Only 15.4% of the participants responded to always having issues with discoloured teeth in comparison to 53.8% never experiencing them. About 69.2% responded never 'had sores or sore spots in or around your mouth' in comparison to 2.6% responding always. In terms of having bad breath and bleeding gums, 53.8% indicated that they never experienced the symptoms, respectively. In terms of 'having food sticking in or between your teeth', 23.1% responded to always having the experience and 25.6% sometimes having the experience. The experience of 'had pain or sensitivity in your teeth with hot or cold stimuli' was low with 30.8% never experiencing the discomfort and 12.8% responding to have always experienced the discomfort. The issue with 'had dry mouth or lips' was also low with 41.0% responding to never having the issue and 10.3% responding with always.

Table 6: Item descriptive statistics for oral symptoms and functional wellbeing

Items	Mean (SD)	Min	Max
Had pain in your teeth/toothache?	2.10 (1.165)	1	5
Been breathing through your mouth or snoring?	2.33 (1.64)	1	5
Had discoloured teeth or spots on your teeth?	2.21 (1.56)	1	5
Had crooked teeth or spaces between the teeth?	3.85 (1.84)	1	6
Had sores or sore spots in or around your mouth?	1.77 (1.51)	1	6
Had bad breath?	1.96 (1.36)	1	6
Had bleeding gums?	2.13 (1.56)	1	6
Had food sticking in or between your teeth	2.97 (1.54)	1	6
Had pain or sensitivity in your teeth with hot or cold stimuli?	2.79 (1.79)	1	6
Had dry mouth or lips?	2.46 (1.70)	1	6

Emotional wellbeing

The mean scores for all emotional wellbeing items (Table 7) were between 2.28 and 3.23, indicating that the participants reported moderate to severe levels of emotional wellbeing problems. An analysis of emotional wellbeing indicated that only 12.8% of the participants felt unhappy or sad because of their teeth, mouth, or face, contrary to 30.8% who responded to never experiencing sadness about their teeth, mouth, or face. Furthermore, 15.4% of participants always felt confident about their teeth, mouth, or face compared to 38.5% who did not have confidence. Concerning the teeth, mouth, or face, 17.9% always felt anxious, shy, or withdrawn and 25.6% felt they were different from others. A total of 30.8% have never felt good looking/attractive due to their teeth, mouth, or face. There was less worry about what other people think with 35.9% of participants highlighting they had never been worried in comparison to 7.7% who always felt worried. About being asked questions about their teeth, mouth, or face, 38.9% never felt upset/uncomfortable in comparison to 17.9% responding to always having an emotional response.

Table 7: Item descriptive statistics for emotional wellbeing

Items	Mean (SD)	Min	Max
Been unhappy or sad because of your teeth, mouth, or face?	2.92 (1.87)	1	6
Been confident because of your teeth, mouth, or face?	2.72 (1.91)	1	6
Felt worried or anxious because of your teeth, mouth, or face?	3.05 (1.91)	1	6
Felt shy or withdrawn because of your teeth, mouth, or face?	2.87 (1.99)	1	6
Felt unattractive because of your teeth, mouth, or face?	2.82 (2.01)	1	6
Been angry because of your teeth, mouth, or face?	2.28 (1.65)	1	6
Felt that you look different because of your teeth, mouth, or face?	3.23 (1.97)	1	6
Felt that you were attractive (good looking) because of your teeth, mouth, or face?	2.74 (1.73)	1	6
Been worried about what other people think or say about your teeth, mouth, or face?	2.97 (2.11)	1	6
Been upset or uncomfortable with being asked questions about your teeth, mouth, or face?	2.67 (1.80)	1	6

Schooling

The mean scores for all schooling items (Table 8) were between 1.59 and 2.67, indicating that the participants reported moderate to severe levels of educational problems. In terms of schooling, most participants (56.4%) responded to never missing school due to any reason because of their teeth, mouth, or face, whereas only 5.1% of respondents always missed school. The majority (59%) never had trouble paying attention in school because of their teeth, mouth, or face, and 41.0% never experienced not wanting to speak/read out loud in class because of their teeth, mouth, or face. Only 7.7% and 12.8% usually had difficulty paying attention and

not wanting to speak/read out loud, respectively due to their teeth, mouth, or face. A total of 82.1% never experienced not wanting to go to school because of their teeth. In terms of being teased or punished by teachers or peers for not being able to articulate speech clearly because of the cleft, 17.9% responded with sometimes having the experience and 15.4% responded with always having the experience. About 41% never experienced being teased or punished by teachers or peers.

Table 8: Item descriptive statistics for schooling

Items	Mean (SD)	Min	Max
Missed school for any reason because of your teeth, mouth, or face?	1.95 (1.49)	1	6
Had difficulty paying attention in school because of your teeth, mouth, or face?	1.87 (1.42)	1	6
Not wanted to speak/read out loud in class because of your teeth, mouth, or face?	2.38 (1.57)	1	6
Not wanting to go to school because of your teeth?	1.59 (1.48)	1	6
Been teased or punished by teachers or peers for not being able to articulate speech clearly because of the cleft?	2.67 (1.84)	1	6

Peer relationship

The results in Table 9 for item descriptive statistics for peer relationship show that the mean score for all items was between 1.87 and 3.10. The highest response to peer relations relates to being asked questions because of their teeth, mouth, or face, and 30.8% of the participants stated they have always been questioned by their peers. This was followed by 12.8% who always do not want to meet new people because of their teeth, mouth, or face. In terms of being teased, bullied, or called names by other children because of their teeth, mouth, or face, 30.8% of the participants sometimes faced bullying. About 15.4% of the participants sometimes avoided talking, smiling, or laughing with other children because of their teeth, mouth, or face in comparison to 56.4% responding to never having the experience. Conflict with friends or family because of their teeth, mouth, or face was low, with 61.5% never experiencing the conflict and 7.7% always experiencing it.

Table 9: Item descriptive statistics for peer relationship

Items	Mean (SD)	Min	Max
Avoided talking, smiling, or laughing with other children because of your teeth, mouth, or face?	2.10 (1.54)	1	6
Been teased, bullied, or called names by other children because of your teeth, mouth, or face?	2.15 (1.56)	1	6
Been asked questions because of your teeth, mouth, or face?	3.10 (1.68)	1	6
Not wanted to meet new people because of your teeth, mouth, or face?	1.97 (1.51)	1	6
Been fighting or arguing with other children or family members because of your teeth, mouth, or face?	1.87 (1.87)	1	6

Family impact

The results in Table 10 for item descriptive statistics for family impact show that most parents believe their child's condition hurts their family. The mean score for all items was between 2.26 and 3.33, with a standard deviation of between 1.84 and 1.98. For family impact it was observed that 23.1% of the guardians always (vs 38.5% responding *never*) experienced that the child's condition caused any financial burden; 20.5% always (vs 30.8% responding *never*) have themselves or other guardian taken time out from work due to the child's condition; and 28.2% always (vs 25.6% responding *sometimes*) experience that their child requires more attention from them. In terms of concerns about the future wellbeing of the child, 10.3% of the guardians always felt concerned, 25.6% sometimes worried, and 30.8% never felt worried.

Most of the guardians (53.8%) never experienced conflict in the family internally with siblings or other family members due to the child's condition, whereas 23.1% would sometimes face conflict. There was high uncertainty (28.2%) about guardians or their partner/family members having concerns about the future wellbeing of the child. This was followed by 23.1% uncertainty concerning the guardian or the other parent feeling guilty for the child's state. About 23.1% of the guardians sometimes respectively experienced conflict in the family internally with siblings or other family members (vs 53.8% responding *never*), felt upset because of the child's condition (vs 43.6% responding *never*), and felt guilty for the child's

state (43.7% responding *never*). Only 15.4% of the guardians and partners sometimes had their sleep affected and 56.4% never had it affected. Concerning feeling uncomfortable in public spaces or being ridiculed or stigmatized when with the child, 59.0% never had the experience and 12.8% sometimes had the experience.

Table 10: Item descriptive statistics for family impact

Items	Mean (SD)	Min	Max
Has the child's condition cause any financial burden?	2.95 (2.04)	1	6
Have you or your partner/family member taken time out from work due to the child's condition?	2.85 (1.84)	1	6
Does your child require more attention from you?	3.33 (1.92)	1	6
Has the child's condition caused conflict in the Family internally with siblings or other family members?	2.26 (1.87)	1	6
Have you felt uncomfortable in public spaces or been ridiculed or stigmatized when with the child?	2.36 (2.01)	1	6
Have you or your partner/family member had concerns about the future wellbeing of the child?	3.21 (2.13)	1	6
Have you or the other parent felt upset because of the child's condition?	2.62 (1.97)	1	6
Have you or the other parent felt guilty for the child's state?	2.64 (2.03)	1	6
Has your sleep or that of your partner been affected?	2.36 (1.98)	1	6

General health

Figure 3 shows the frequency distribution of the general health of participants with OFC. The mean general health score was 4.44, with a standard deviation of 0.821. This means that the average participant's general health was good, but there was a range of scores, with some participants reporting poorer or better health. The most common general health score was 4(*good*), followed by 5 (*great*). There were fewer participants with lower general health scores.

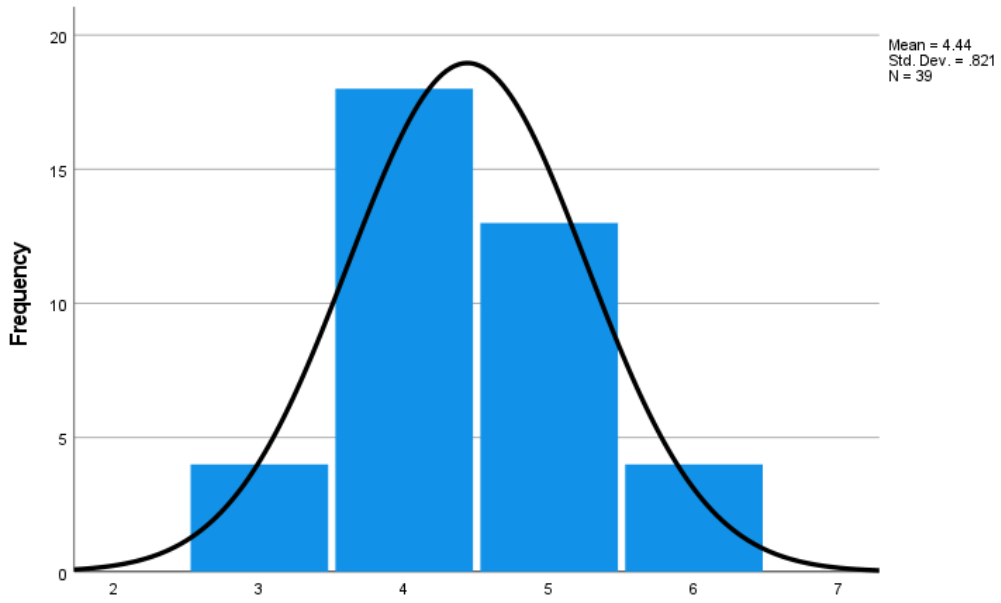

Figure 3*: Distribution of general health among participants with OFC*

Discussion

This investigation examined the oral health related quality of life (OHRQoL) among 39 individuals with orofacial clefts (OFC). The demographic profile of the sample predominantly comprised young individuals, with 71.8% below the age of 18. The age of presentation for OFC at hospital is usually under the age of 1 year. The observed age distribution may reflect improvements in healthcare and health education systems. Because OFC causes infants to have difficulty feeding, their parents are likely to bring them to the hospital earlier. Among various OFC types, patients with isolated cleft palate presented earlier than those with isolated cleft lip.

Gender distribution highlighted that male constituted most participants, accounting for 56.4%. Cleft lip and palate emerged as the most prevalent OFC type, affecting 61.5% of the participants. These findings regarding gender distribution and cleft type align with prior studies, which have noted a higher occurrence of OFC, particularly cleft lip, and palate, among males compared to females. Conversely, males typically exhibit a higher prevalence of isolated cleft palate (Adekeye and Lavery, 1985, Al Omari and Al-Omari, 2004, Noorollahian et al., 2015).

The overall OHRQoL among the participants was generally favourable, as evidenced by a mean score of 198.12. However, certain aspects of their OHRQoL exhibited areas for potential enhancement. Notably, participants reported experiencing moderate to severe levels of oral symptoms and functional well-being challenges, emotional well-being concerns, and educational difficulties. Lower functional well-being scores observed in teenagers may be attributed to their experiencing untreated disease for a longer duration compared to the younger participants (Broder and Wilson-Genderson, 2007)

It has been highlighted that children with CL/P have a significantly poorer OHRQoL compared to their peers without CL/P, with social-emotional well-being greatly impacted during teenage years (Ward et al., 2013, Kortelainen et al., 2016). It is noteworthy that the participants experienced OHRQoL differences in all the aspects of OHRQoL, measured in the study, for instance, the overall oral health, the symptoms experienced, the functional limitations, emotional welfare, and social welfare.

Non-syndromic CL/P patients experience lower OHRQoL compared to the general non-cleft population (Antonarakis et al., 2013). Factors contributing to low OHRQoL include variations within cleft subgroups, patient satisfaction with treatment, ethnicity, access to healthcare, and surgical recommendations. Cleft lip and/or palate impacts oral health, social interactions, and overall quality of life. Research suggests that dissatisfaction with post-cleft treatment aesthetics can contribute to a diminished QoL for patients (Oosterkamp et al., 2007, Gkantidis et al., 2013). This is evidenced by the connection between perceived facial imperfections and reduced social and professional engagement. The findings emphasize the importance of achieving the most optimal surgical outcomes for individuals with clefts.

Our study underscores the need for comprehensive care in OFC patients, addressing their physical, emotional, and social well-being. Instruments like the Child Perceptions Questionnaire (CPQ) exemplify this approach by assessing these key domains (Jokovic et al., 2003). Furthermore, successful cross-cultural adaptations of the CPQ pave the way for culturally sensitive interventions tailored to the unique experiences of individuals with OFC (Wogelius et al., 2009). Importantly, research by Broder and co-workers (2012) demonstrated the feasibility of developing shorter, reliable, and valid instruments such as the Child Oral Health Impact Profile-Short Form (COHIP-SF) 19. The COHIP-SF19 offers a significant advantage in streamlining clinical research and epidemiological studies involving children (Broder et al., 2012). Notably, all the questionnaires demonstrate high reliability through Cronbach's alpha coefficients exceeding 0.8. Similarly to our study findings Cronbach's alpha of 0.898 which demonstrated high reliability.

In a study evaluating well-being (QoL) in relation to mouth problems (oral symptoms) and ability to perform daily activities (functional wellbeing), nearly 38.5% of participants reported never experiencing tooth discomfort. However, 30.8% indicated occasional tooth pain. Breathing difficulties, stained teeth, mouth ulcers, halitosis, bleeding gums, difficulty removing food particles, and sensitivity to temperature changes were frequently reported. A study on children with and without CL/P has shown that children with CL/P have more central apnea, which suggests a problem with their breathing control system, compared to obstructive apnea which are more common and caused by a blocked airway (Maclean et al., 2017). A study found that adolescents with cleft lip or palate face various oral health issues, including tooth pain, decay, developmental delays, missing teeth, cosmetic concerns, gum problems, mouth ulcers, bad breath, and facial deformities which affected their QoL (Pisek et al., 2014).

Oral symptoms and functional wellbeing have a significant correlation with both emotional wellbeing and peer relationship. In terms of emotional wellbeing, our study findings showed that only 12.8% of participants experienced sadness or sadness due to their teeth, mouth, or face, while 15.4% felt confident. Higher prevalence concerning mental health diagnoses among adults with CL/P. Nearly half (45.9%) reported having a diagnosed condition (Ardouin et al., 2020).

According to the literature, individuals with OFC have a higher prevalence of depression than the general population (Pedersen et al., 2016, Xia et al., 2023). Additionally, self-reported rates of post-traumatic stress disorder (PTSD) and eating disorders were also slightly elevated (Ardouin et al., 2020). Our study shows that emotional wellbeing is greatly influenced by aesthetics associated with OFC. These findings align with other prior research suggesting increased risk for depression, anxiety, and poorer overall emotional well-being in individuals with CL/P. Several factors may obscure the psychosocial well-being of children with CL/P, including difficulties related to coping and adjustment, self-confidence, bullying, facial appearance acceptance, surgical satisfaction, communication, parental anxiety and depression, and financial strain (Al-Namankany and Alhubaishi, 2018). Whilst children with CL/P may generally exhibit psychological outcomes comparable to the unaffected population, certain symptoms, including anxiety and depression, seem to be more prevalent in this group (Branson et al., 2022).

The current study found that 30.8% of participants were always questioned by their peers due to their teeth, mouth, or face, while 12.8% disliked meeting new people. 30.8% experienced bullying, while 15.4% avoided talking or laughing with others. Conflict with friends or family was low, with 61.5% never experiencing it and 7.7% always experiencing it. This highlights the need for psychosocial support for OFC patients to deal with peer and family relationships. Research emphasizes the significance of ongoing psychosocial support from a young age for individuals with CL/P (Ardouin et al., 2021). This support can facilitate the development of social skills, self-esteem, and social confidence, ultimately fostering fulfilling friendships and romantic relationships. The study on children with craniofacial anomalies (CFA) found that they value friendships and find them easy to establish (Pope et al., 2016). Mothers play a crucial role in providing support and guidance (Pope et al., 2016).

Our study findings for the school-aged participants, the results showed that most participants (56.4%) never missed school due to their teeth, mouth, or face, and 59% never had trouble paying attention or speaking out loud. Over eighty-two percent never experienced not wanting to go to school. Children with CL/P are at high risk of experiencing otitis media (ear infections) or middle ear effusion (fluid buildup) by the age of seven (Goh et al., 2019). This can lead to hearing problems that may negatively impact their social interactions with peers and academic performance in school. Research investigating teachers' perceptions of CL/P underscored the critical role of knowledgeable and supportive school environments for students with health conditions (Stock et al., 2018). Some of the participants in our study had their schooling attendance affected, and thus teachers can play a role in ensuring the students are well accommodated within the classroom. The study further emphasized the need for teachers to receive training on CL/P treatment protocols and social support strategies. A multidisciplinary treatment approach was shown to significantly improve speech and language difficulties in young children with clefts, bringing their abilities closer to those of unaffected peers (Ruiter et al., 2009).

A notable difference between this study and previous research is its focus on individuals with OFC in South Africa. South Africa is a region with distinct healthcare settings, cultural norms, and socioeconomic factors that can influence OFC experiences (Williams et al., 2016). Previous studies have often focused on individuals from developed countries, potentially overlooking the unique challenges faced by those in resource-limited settings (Defabianis et al., 2022). By including participants from South Africa, this study provides valuable insights into the contextual factors that shape OFC's impact and the importance of culturally sensitive interventions.

The study findings also highlighted the potential for conflict within families due to OFC. The current study findings regarding the family impact of children with OFCs have identified financial strain, work leave, and heightened caregiving demands. Although concerns about the child's future were common, familial conflict appeared less frequent. Guardians reported some uncertainty regarding guilt and sleep disruption, albeit to a lesser degree than the burdens. From a parent/guardian perception towards their child's OFC condition, a study by Hunt and colleagues concluded that parents of children with CL/P believe their children experience psychosocial challenges, including lower self-esteem, higher anxiety, more behavioural problems, and dissatisfaction with speech compared to children without CL/P (Hunt et al.,

2007). Our study highlights that guardians/parents' mental health is also crucial as their QoL can be affected.

While over half (59.0%) of participants in the current study reported no experiences of public stigma or ridicule when with their child, a study in Nigeria has documented higher rates of stigma (Adeyemo et al., 2016). This disparity highlights the influence of cultural beliefs, as the Nigerian study linked societal misconceptions about CL/P to increased stigma for affected individuals and families. The findings underscore the importance of community-based interventions to address these misconceptions and raise awareness about CL/P causes, treatments, and support resources. In South Africa, various conceptions exist regarding the aetiology of OFCs, inter alia beliefs in consanguinity (blood relations between parents), internal foetal abnormalities, divine punishment, natural occurrence, dietary influences during pregnancy, physical trauma during pregnancy, teratogenic medication use, and interpretations of OFCs as God's creation or gift (Penn et al., 2009).

Like Lentge et al. (2022) who have acknowledged the emotional stress experienced by parents of children with OFC, this study extends the understanding of the possibility of conflicts between siblings and other members of the family (Lentge et al., 2022). About 7.7% our participants reported to always been fighting or arguing with other children or family members because of your teeth, mouth, or face. High levels of family solidarity and openness and less conflict with family members typically give rise to these positive characteristics: self-confidence, good social skills, and psychological health among youths and young adults. A study has highlighted that mothers and fathers of children with oral clefts experience psychosocial adjustment differently (Nidey et al., 2016). Mothers may face a greater burden, potentially employing coping mechanisms such as self-blame, emotional release, and acceptance (Berger and Dalton, 2011). Conversely, fathers demonstrated higher levels of resilience compared to mothers (Ueki et al., 2019). This resilience manifested in stronger problem-solving abilities and a greater capacity for recognition and acceptance. Additionally, parents from lower socioeconomic backgrounds appear to be at higher risk for these burdens (Nidey et al., 2016). These factors can lead to observed conflicts that have been reported in our study.

The novelty of our study lies in its comprehensive assessment of OFC's impact on various aspects of quality of life, its inclusion of younger participants, and its identification of potential family conflicts. These findings provide valuable insights for healthcare providers,

policymakers, and researchers working to improve the lives of individuals with OFC and their families. These findings underscore the need for family-based interventions that address not only the individual with OFC but also the broader family system.

An average health welfare of 4.44 was observed in our study from the parents/guardians' viewpoint on the child's wellbeing, which indicated good overall health. The age of the child may influence the cognition of the parent when trying to assess the health of the child. Cleft palate in younger children has been shown to lead to reduced food intake and weight gain, causing concern for parents due to insufficient suction and regurgitation (De Cuyper et al., 2019). Psychosocial functioning reporting for parents of children with CL/P usually report the child having lower self-esteem, anxiety, and overall unhappiness compared to those without CL/P, as well as more behavioural issues and less speech satisfaction (Hunt et al., 2007). A study on parental perspectives underscored the ongoing need for empathy, support, and access to information at every stage of their child's journey (Stone et al., 2010). These stages were identified as the time of diagnosis at birth, during surgical procedures (especially the initial surgery), and upon school entry (Stone et al., 2010).

The findings of our study have several important clinical implications for the care of individuals with OFC. Early and comprehensive assessments of various aspects of quality of life, including oral health, emotional wellbeing, social functioning, and educational experiences, have been reported as crucial to identifying potential challenges and providing timely interventions (Khanchezar et al., 2019). Addressing the impact of physical anomalies on oral functions, speech production, and overall appearance requires a collaborative approach among dentists, orthodontists, speech pathologists, and surgeons (Khanchezar et al., 2019).

The limitations of this study include its cross-sectional design, which means that it cannot prove cause and effect. Additionally, the study did not assess the impact of specific treatments on quality of life. A cross-sectional design can only establish associations between variables, not causal relationships. Longitudinal studies are needed to determine the long-term impact of OFCs on quality of life and to assess the effectiveness of specific treatments. This study did not assess the impact of specific treatments on quality of life. Future research should investigate the effectiveness of different interventions, including medical, dental, psychological, and social support, in improving quality of life for individuals with OFCs.

Conclusions and Recommendations

Conclusions

This study examining the OHRQoL in children with OFCs treated at a university hospital revealed several significant conclusions and implications. It unequivocally confirmed that OFCs have a profound impact on the OHRQoL of affected children. These congenital conditions introduce a range of physical and psychological challenges, including pain, difficulties with eating and speaking, and emotional distress. These challenges collectively diminish the overall quality of life for these children.

One striking observation from the findings was the deficiency in orthodontic care as a substantial portion of the participants did not receive orthodontic braces. The absence of such interventions was correlated with poorer OHRQoL outcomes. This underscores the critical importance of ensuring comprehensive care, which includes orthodontic treatment to enhance the wellbeing of children living with OFCs.

The findings also suggested that the severity of the cleft condition significantly influences OHRQoL. Those with more severe conditions, such as cleft lip and palate, experience heightened physical and psychological challenges. Thus, addressing the unique needs of children with more severe cleft conditions is imperative to improve their quality of life.

Access to specialised care emerged as a crucial factor in determining the OHRQoL of these children. Enhancing access to specialised care and ensuring timely interventions can substantially improve their overall quality of life. Moreover, socioeconomic factors were identified as an additional layer of complexity, with children from disadvantaged backgrounds facing additional barriers to care and support. Addressing these socioeconomic disparities is pivotal to achieve better outcomes.

The findings also shed light on the burden faced by caregivers of children with OFCs who reported experiencing stress, overwhelm, and a lack of support. This highlights the necessity of providing psychosocial support and resources to caregivers to enable them to better assist their children.

Recommendations

Considering these findings, the study recommends a holistic approach to enhance the OHRQoL of children with OFCs. Such an approach should encompass not only medical and orthodontic interventions but also comprehensive psychosocial support for both the affected children and their caregivers. Developing care plans that address the physical, psychological, and social aspects of the condition is essential to improving their quality of life.

Furthermore, these conclusions have broader implications for healthcare policies and practices. Policymakers and healthcare providers should prioritise early intervention, improve access to orthodontic treatment, and provide psychosocial support to children with OFCs. This comprehensive patient-centred approach is fundamental to enhancing the overall wellbeing of these children. We suggest carrying out more research at other facilities that treat CL/P throughout South Africa to get a full nationwide view. It is also crucial to address disparities in access to care and support, particularly for children from different socioeconomic backgrounds, to ensure equitable outcomes.

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Appendices

Appendix 1: Oral Health-Related Quality of Life Questionnaire

Dear Participant

Thank you for agreeing to take part in the study. I am doing this study to better understand how children and parents feel about their cleft condition and about themselves.

Please carefully read each statement and choose the answer that best describes you in the past 3 months regarding your cleft condition, mouth, face, and schooling. There are no right or wrong answers. We want to know how you really feel.

Example:

During the **past 3 months**, how often have you **felt shy** because of your teeth, mouth, or face?

If you have felt shy because of your teeth, mouth, or face then choose the appropriate response listed below "Never, Sometimes, Regularly, Usually, Always or Uncertain". If you felt shy for other reason, please skip to the next question.

Never	**Sometimes**	**Regularly**	**Usually**	**Always**	**Uncertain**
□ (1)	□ (2)	□ (3)	□ (4)	□ (5)	□ (6)

Please remember:

- To answer the questions as openly as you can.
- Not to talk to anyone about the questions when you are answering them.
- Before you are answering, ask yourself: Does this happen because of the cleft condition.
- Choose the answer that best describes you in the past 3 months.
- Start answering the questions in the next page.

Oral Health Related Quality of Life Questionnaire

A. BIOGRAPHIC INFORMATION

(Section A-E to be answered by patient/child with CL/P)

What is your gender? □Male □ Female □ Other

Race? □ Black □ Coloured □ Indian □ White □ Other

How old are you? _____years

What type of cleft do you have? □ Cleft lip □ Cleft palate □ Cleft lip and palate

Do you have braces? □ Yes □ No

B. ORAL SYMPTOMS AND FUNCTIONAL WELL BEING

In the past 3 months have you had or experienced any of the following oral symptoms	**1**	**2**	**3**	**4**	**5**	**6**
1. Had pain in your teeth/toothache?						
2. Been breathing through your mouth or snoring?						
3. Had discoloured teeth or spots on your teeth?						
4. Had crooked teeth or spaces between the teeth?						
5. Had sores or sore spots in or around your mouth?						
6. Had bad breath?						
7. Had bleeding gums?						
8. Had food sticking in or between your teeth						
9. Had pain or sensitivity in your teeth with hot or cold stimuli?						
10. Had dry mouth or lips?						

C. EMOTIONAL WELL-BEING

In the past 3 months have you had or experienced any of the following regarding your emotional well-being?	**1**	**2**	**3**	**4**	**5**	**6**
1. Been unhappy or sad because of your teeth, mouth, or face?						
2. Been confident because of your teeth, mouth, or face?						
3. Felt worried or anxious because of your teeth, mouth, or face?						
4. Felt shy or withdrawn because of your teeth, mouth, or face?						
5. Felt unattractive because of your teeth, mouth, or face?						
6.Been angry because of your teeth, mouth, or face?						
7. Felt that you look different because of your teeth, mouth, or face?						
8. Felt that you were attractive (good looking) because of your teeth, mouth, or face?						
9. Been worried about what other people think or say about your teeth, mouth, or face?						
10. Been upset or uncomfortable with being asked questions about your teeth, mouth, or face?						

D. SCHOOLING

In the past 3 months have you had or experienced any of the following regarding school	1	2	3	4	5	6
1. Missed school for any reason because of your teeth, mouth, or face?						
2. Had difficulty paying attention in school because of your teeth, mouth, or face?						
3. Not wanted to speak/read out loud in class because of your teeth, mouth, or face?						
4. Not wanting to go to school because of your teeth?						
5. Been teased or punished by teachers or peers for not being able to articulate speech clearly because of the cleft?						

E. PEER RELATIONSHIP

In the past 3 months how can you describe your experience regarding the relationship with peers	1	2	3	4	5	6
1. Avoided talking, smiling, or laughing with other children because of your teeth, mouth, or face?						
2.Been teased, bullied, or called names by other children because of your teeth, mouth, or face?						
3. Been asked questions because of your teeth, mouth, or face?						
4. Not wanted to meet new people because of your teeth, mouth, or face?						
5. Been fighting or arguing with other children or family members because of your teeth, mouth, or face?						

F. FAMILY IMPACT (To be answered by Parent/Guardian of child with CL/P)

In the past 3 months how can you describe the relationship of the child and the family	1	2	3	4	5	6
1. Has the child condition cause any financial burden?						
2. Have you or your partner/family member taken time out from work due to the child's condition?						
3. Does your child require more attention from you?						
4. Has the child's condition caused conflict in the Family internally with siblings or other family members?						
5. Have you felt uncomfortable in public spaces or been ridiculed or stigmatized when with the child?						
6. Have you or your partner/family member had concerns about the future wellbeing of the child?						
7. Have you or the other parent felt upset because of the child's condition?						
8. Have you or the other parent felt guilty for the child's state?						
9. Has your sleep or that of your partner been affected?						

G. GENERAL HEALTH (To be answered by Parent/Guardian of child with CL/P)

What is the general health of the child?

Bad	Poor	Average	Good	Great	Uncertain
□ (1)	□ (2)	□ (3)	□ (4)	□ (5)	□(6)

_____

_____

_____

_____

END OF FORM - THANK YOU

Appendix 2: Ethical Clearance Certificate

Appendix 3: Permission Letter from the CEO at WOHC

UNIVERSITY OF THE WITWATERSRAND, JOHANNESBURG

R49 Drs T Makhetha

HUMAN RESEARCH ETHICS COMMITTEE (MEDICAL)
CLEARANCE CERTIFICATE NO. M210816

NAME: Dr T Makhetha **DEGREE:** MSc (Dent)
(Principal and Co-Investigators)

DEPARTMENT: School of Oral Health Sciences
Department of Orthodontics
Dental School
University

PROJECT TITLE: *Health-related quality of life in children with orofacial clefts, treated at a university hospital*

DATE CONSIDERED: 27 August 2021

DECISION: Approved unconditionally

CONDITIONS: Change of supervisor noted on 20 March 2024

NOTE: If contact information regarding student study participants is required, please contact the Registrar's office <Niccleen.Potgieter@wits.ac.za>

SUPERVISOR: Professor P Hlongwa and Dr ME Makofane

APPROVED BY:
Professor P Ruff, Chairperson, HREC (Medical)

DATE OF APPROVAL: 21 February 2022 **EXPIRY DATE:** 20 February 2027

This Clearance Certificate is valid for 5 years from the date of approval. An extension may be applied for.

DECLARATION OF INVESTIGATORS

To be completed in duplicate and **ONE COPY** returned to the Research Office secretariat on the 3rd floor, Phillip Tobias Building, Parktown, University of the Witwatersrand, Johannesburg.

I/we fully understand the conditions under which I am/we are authorized to carry out the above-mentioned research and I/we undertake to ensure compliance with these conditions. Should any departure be contemplated from the research protocol as approved, I/we undertake to submit details to the Committee. **I agree to submit a yearly progress report** in the format available at https://wits.ac.za/research/researcher-support/research-ethics/ethics-committees/. This report is due annually, for the duration of the Clearance Certificate, beginning on the first anniversary of Date of Approval above. Unreported changes to the study may invalidate the clearance given by the HREC (Medical).

Signature of Principal Investigator

21/02/2022
Date

Appendix 4: Participants' Information Sheet: Parents/caregivers

Study title: Health-Related Quality of Life in Children with Cleft Lip and Palate who are treated at a University Hospital.

Introduction

I am Dr T Makhetha, a postgraduate student at the University of the Witwatersrand. I am doing research for my Master's degree on Health-Related Quality of Life in Children with Cleft Lip and Palate (CL/P) who are treated at Wits Oral Health Centre (WOHC).

I am going to give you information and invite you to be part of this research. If there is any information that you do not understand, please ask me to stop as we go through the information, and I will take time to explain. If you have questions later, you are welcome to ask them at any time.

Purpose of the research

The study will be conducted at the Cleft Clinic of the WOHC. Cleft lip and palate patients receiving orthodontic treatment will be recruited to participate in the study. The purpose of the study is to check how orofacial cleft affects the health and the way of life in children born with CL/P.

Type of Research Intervention

This research will involve your participation to complete the questionnaire if your child has cleft lip and/or palate and receiving orthodontic treatment.

Participant Selection

You are being invited to take part in this research because we feel that your child's experience as a cleft lip and palate patient can contribute much to our understanding and knowledge of how orofacial cleft affects your child's way of life.

Voluntary Participation

Your participation in this research is entirely voluntary. It is your choice whether to participate or not. If you choose not to participate all the services, your child is receiving at WOHC will.

continue and nothing will change.

Procedures

You will be asked to fill out a survey which will be provided. In the survey there is a section specifically for parents/caregivers. There is a separate section for children. The children will only be able to take part with the prior consent of their parent or guardian. The questions will be asked to you away from your child for your comfort to allow you to be able to answer the questions as openly as possible. You may answer the questionnaire yourself, or it can be read to you, and you can say out loud the answer you want me to write down. If you do not wish to answer any of the questions included in the survey, you may skip them and move on to the next question. The information recorded is confidential, your name is not being included on the forms, and no one else except principal investigators (Dr T Makhetha) will have access to your survey.

Duration

The questionnaire will take 15 minutes to complete.

Risks

There are no anticipated risks in this study.

Benefits

There will be no direct benefit to you, but your participation might help us understand more about how cleft lip and palate condition affects the daily life of your child.

Reimbursements

There is neither cost nor payment for taking part in the research.

Confidentiality

All information taken from the study will be coded to protect each participant's name. No names or other identifying information will be used when discussing or reporting data. The investigator(s) will safely keep all files and data collected in a secured locked cabinet in the principal investigator's office.

Sharing the Results

Nothing that you tell us today will be shared with anybody outside the research team, and nothing will be attributed to you by name. The data will only be combined with other participant's data to generate a statistical analysis which can be published and presented in research journals, discussions, or seminars without participant's information.

I will be pleased to provide you with a results summary on request, at no cost.

Right to Refuse or Withdraw

You do not have to take part in this research if you do not wish to do so and choosing to participate or not will not affect your child's treatment in any way.

Who to Contact?

If you have any questions, you can ask them now or later. If you wish to ask questions later, you may contact any of the following:

Dr T Makhetha: Principal Investigator, Cell: 0781919146, Email: 723815@students.wits.ac.za

Dr M Moshaoa: Supervisor, Tel No. 011 488 4850, Email: Mpule.Moshaoa@wits.ac.za

This study has been approved by the Human Research Ethics Committee (Medical) of the University of the Witwatersrand, Johannesburg ("Committee"). A principal function of this Committee is to safeguard the rights and dignity of all human subjects who agree to participate in a research project and the integrity of the research.

If you have any concern over the way the study is being conducted, please contact the Chairperson of this Committee who is Dr Clement Penny, who may be contacted on telephone number 011 717 2301, or by e-mail on Clement.Penny@wits.ac.za. The telephone numbers for the Committee secretariat are 011 717 2700/1234 and the e-mail addresses are Zanele.Ndlovu@wits.ac.za and Rhulani.Mukansi@wits.ac.za

Thank you for reading this Study Information Sheet.

March 2022

Appendix 5: Participants' Information Sheet: Children Under 18 Years of Age

Study title: Health Related Quality of Life in Children with Cleft Lip and Palate who are treated at a University Hospital.

Introduction

I am Dr T Makhetha, a postgraduate student at the University of the Witwatersrand. I am doing research on Health-Related Quality of Life in Children with Cleft Lip and Palate (CL/P) who are treated at Wits Oral Health Centre (WOHC). You are one of the children who has been getting treatment for this condition.

I am going to give you information and invite you to be part of this research. If there is any information that you do not understand, please ask me to stop as we go through the information, and I will take time to explain. If you have questions later, you are welcomed to ask them at any time.

Why are we doing the research?

The study will be conducted at the Cleft Clinic of the WOHC. Cleft lip and palate patients receiving orthodontic treatment will be recruited to participate in the study. The purpose of the study is to check how orofacial cleft affects the health and the way of life in children born with CL/P.

How will you participate in the research?

This research will involve your participation in completing a questionnaire if you have cleft lip and/or palate and receiving orthodontic treatment.

How are you chosen to be part of the research?

You are being invited to take part in this research because we feel that your experience of living with cleft lip and palate can contribute much to our understanding and knowledge of how orofacial cleft affects your way of life.

Free participation

Your participation in this research is entirely voluntary. It is your choice whether to participate or not. If you choose not to participate all the services that you receive at WOHC will continue and not.

Ways of answering the questions

You will be asked to fill out part of a questionnaire which will be provided in a quiet place in the clinic. You may answer the questionnaire yourself, or it can be read to you, and you can say out loud the answer you want me to write down. Your parent/caregiver may also assist to answer the questions for you. If you do not want to answer any of the questions included in the survey, you may skip them and move on to the next question. Your parent or guardian will separately be asked to fill out another part of the questionnaire. The information recorded is confidential, your name is not being written on the forms, and no one else except me (Dr T Mahketha), will have access to your answers.

Permission

Both you and your parent or guardian will have to agree that you can take part.

Duration

The questionnaire will take 15 minutes to complete.

Risks

There are no anticipated risks in this study.

Benefits

There will be no direct benefit to you, but your participation might help us understand more about how cleft lip and palate condition affects your daily living.

Reimbursements

There is neither cost nor payment involved in taking part in the study.

Confidentiality

All information taken from the study will be coded to protect each participant's name. No names or other identifying information will be used when discussing or reporting data. The investigator(s) will safely keep all files and data collected in a secured locked cabinet in the principal investigator's office.

Sharing the Results

Nothing that you tell us today will be shared with anybody outside the research team, and nothing will be attributed to you by name. The data will only be combined with other participant's data to generate a statistical analysis which can be published and presented in research journals, discussions, or seminars without participant's information.

I will be pleased to provide you with a results summary on request, at no cost.

Right to Refuse or Withdraw

You do not have to take part in this research if you do not wish to do so and choosing to participate or not will not affect treatment in any way.

Who to Contact?

If you have any questions, you can ask them now or later. If you wish to ask questions later, you may contact any of the following:

Dr T Makhetha: Principal Investigator, Cell: 0781919146, Email: 723815@students.wits.ac.za

Dr M Moshaoa: Supervisor, Tel No. 011 488 4850, Email: Mpule.Moshaoa@wits.ac.za

This study has been approved by the Human Research Ethics Committee (Medical) of the University of the Witwatersrand, Johannesburg ("Committee"). A principal function of this Committee is to safeguard the rights and dignity of all human subjects who agree to participate in a research project and the integrity of the research.

If you have any concern over the way the study is being conducted, please contact the Chairperson of this Committee who is Dr Clement Penny, who may be contacted on telephone number 011 717 2301, or by e-mail on Clement.Penny@wits.ac.za. The telephone numbers for the Committee secretariat are 011 717 2700/1234 and the e-mail addresses are Zanele.Ndlovu@wits.ac.za and Rhulani.Mukansi@wits.ac.za

Thank you for reading this Study Information Sheet.

March 2022

Appendix 6: Consent Form: Adult participants/parents/caregivers

Title: Health Related Quality of Life in Children with Cleft Lip and Palate treated at a University Hospital.

Principle Investigator

Affiliation and Contact Information: Dr. T Makhetha; BDS

Cel:0781919146

Email: 723815@students.wits.ac.za

Consent Form

I have been invited to participate in research titled: -**Health Related Quality of Life in Children with Cleft Lip and Palate treated at a University Hospital.**

I have read the information sheet, (or it has been read to me). I have had the opportunity to ask questions about it and any questions I asked have been answered to my satisfaction. I consent voluntarily to be a participant in this study.

Print Name of Participant_____

Signature of Participant _____

Date _____

Day/month/year

If illiterate [1]

I have witnessed the accurate reading of the consent form to the potential participant, and the individual has had the opportunity to ask questions. I confirm that the individual has given consent freely.

Print name of witness_____ Thumb print of participant

Signature of witness _____Date _____

Statement by the researcher/person taking consent.

I have accurately read out the information sheet to the potential participant, and to the best of my ability. The participant was given an opportunity to ask questions about the study, and all the questions asked by the participant have been answered correctly and to the best of my ability. I confirm that the individual has not been coerced into giving consent, and the consent has been given freely and voluntarily.

A copy of this informed consent form has been provided to the participant.

Print Name of Researcher/person taking the consent_____

Signature of Researcher /person taking the consent_____

Date _____

Day/month/year

[1] A literate witness must sign (if possible, this person should be selected by the participant and should have no connection to the research team). Participants who are illiterate should include their thumb print as well.

* Defined as being persons under the age of 18 (Strode et al.,2011)

Appendix 7: Children Assent Form



PARTICIPANT ASSENT SHEET FOR MINORS*

Topic: Health Related Quality of Life in Children with Cleft Lip and Palate treated at a University Hospital.

1. The study was explained to me, and I understand what will happen if I take part.
2. I was given time to ask any questions I wanted to and was happy with the answers I was given.
3. I understand that I will not benefit from the study, should I agree to take part. I also understand that I will not be paid to take part in the study; taking part will not cost me anything either.
4. I have been given a range of contact details, repeated below, should I require further information at a later stage, or have any cause for concern over anything which is done to me during the study; and
5. I understand that even if I agree to take part in the study, I can change my mind later and stop being a part of the study.
6. My parent(s) or guardian(s) know that I have been invited to take part in the study. They agree that I may do so, but the decision to take part is also mine.

Minors*

Name of Participant: _____

Date: **_____**

Place: **_____**

Signature or mark _____

Name of Parent or Guardian: _____

Date: **_____**

Place: _____

Signature or mark _____

Witnessed by:

Name of Witness: _____

Signature: _____

Date: _____

* Defined as being persons under the age of 18 (Strode et al.,2011)

Appendix 8: OHRQoL scores

Oral Symptoms and Functional Wellbeing						
In the past 3 months have you had or experienced any of the following oral symptoms	**Never n (%)**	**Sometimes n (%)**	**Regularly n (%)**	**Usually n (%)**	**Always n (%)**	**Uncertain n (%)**
1. Had pain in your teeth/toothache?	15 (38.5)	12 (30.8)	7 (17.9)	3 (7.7)	2 (5.1)	0 (0.0)
2. Been breathing through your mouth or snoring?	20 (51.3)	5 (12.8)	3 (7.7)	3 (7.7)	8 (20.51)	0 (0.0)
3. Had discoloured teeth or spots on your teeth?	21 (53.8)	5 (12.8)	3 (7.7)	4 (10.3)	6 (15.4)	0 (0.0)
4. Had crooked teeth or spaces between the teeth?	9 (23.1)	2 (5.1)	1 (2.6)	8 (20.5)	12 (30.8)	7 (17.9)
5. Had sores or sore spots in or around your mouth?	27 (69.2)	6 (15.4)	1 (2.6)	1 (2.6)	1 (2.6)	3 (7.7)
6. Had bad breath?	21 (53.8)	9 (23.1)	3 (7.7)	3 (7.7)	2 (5.1)	1 (2.6)
7. Had bleeding gums?	21 (53.8)	6 (15.4)	5 (12.8)	2 (5.1)	3 (7.7)	2 (5.1)
8. Had food sticking in or between your teeth	8 (20.5)	10 (25.6)	7 (17.9)	4 (10.3)	9 (23.1)	1 (2.6).
9. Had pain or sensitivity in your teeth with hot or cold stimuli?	12 (30.8)	11 (28.2)	2 (5.1)	5 (12.8)	5 (12.8)	4 (10.3)
10. Had dry mouth or lips?	16 (41.0)	10 (25.6)	2 (5.1)	4 (10.3)	4 (10.3)	3 (7.7)
Emotional Wellbeing Response						
In the past 3 months have you had or experienced any of the following regarding your emotional well-being?	**Never n (%)**	**Sometimes n (%)**	**Regularly n (%)**	**Usually n (%)**	**Always n (%)**	**Uncertain n (%)**
1. Been unhappy or sad because of your teeth, mouth, or face?	12 (30.8)	9 (23.1)	5 (12.8)	2 (5.1)	5 (12.8)	6 (15.4)
2. Been confident because of your teeth, mouth, or face?	15 (38.5)	10 (25.6)	1 (2.6)	2 (5.1)	6 (15.4)	5 (12.8)
3. Felt worried or anxious because of your teeth, mouth, or face?	11 (28.2)	10 (25.6)	3 (7.7)	2 (5.1)	7 (17.9)	6 (15.4)
4. Felt shy or withdrawn because of your teeth, mouth, or face?	15 (38.5)	8 (20.5)	2 (5.1)	1 (2.6)	7 (17.9)	6 (15.4)
5. Felt unattractive because of your teeth, mouth, or face?	17 (43.6)	6 (15.4)	1 (2.6)	3 (2.6)	6 (15.4)	6 (15.4)
6.Been angry because of your teeth, mouth, or face?	20 (51.3)	5 (12.8)	6 (15.4)	1 (2.6)	5 (12.8)	2 (5.1)
7. Felt that you look different because of your teeth, mouth, or face?	11 (28.2)	9 (23.1)	1 (2.6)	2 (5.1)	10 (25.6)	6 (15.4)
8. Felt that you were attractive (good looking) because of your teeth, mouth, or face?	12 (30.8)	11 (28.2)	4 (10.3)	2 (5.1)	7 (17.9)	3 (7.7)
9. Been worried about what other people think or say about your teeth, mouth, or face?	14 (35.9)	10 (25.6)	1 (2.6)	1 (2.6)	3 (7.7)	10 (25.6)
10. Been upset or uncomfortable with being asked questions about your teeth, mouth, or face?	15 (38.5)	9 (23.1)	2 (5.1)	3 (7.7)	7 (17.9)	3 (7.7)

Oral Symptoms and Functional Wellbeing						
In the past 3 months have you had or experienced any of the following oral symptoms	**Never n (%)**	**Sometimes n (%)**	**Regularly n (%)**	**Usually n (%)**	**Always n (%)**	**Uncertain n (%)**
Schooling Responses						
In the past 3 months have you had or experienced any of the following regarding school	**Never n (%)**	**Sometimes n (%)**	**Regularly n (%)**	**Usually n (%)**	**Always n (%)**	**Uncertain n (%)**
1. Missed school for any reason because of your teeth, mouth, or face?	22 (56.4)	10 (25.6)	0 (0.0)	3 (7.7)	2 (5.1)	2 (5.1)
2. Had difficulty paying attention in school because of your teeth, mouth, or face?	23 (59.0)	9 (23.1)	1 (2.6)	3 (7.7)	1 (2.6)	2 (5.1)
3. Not wanted to speak/read out loud in class because of your teeth, mouth, or face?	16 (41.0)	9 (23.1)	4 (10.3)	5 (12.8)	3 (7.7)	2 (5.1)
4. Not wanting to go to school because of your teeth?	32 (82.1)	2 (5.1)	1 (2.6)	0 (0.0)	1 (2.6)	3 (7.7)
5. Been teased or punished by teachers or peers for not being able to articulate speech clearly because of the cleft?	16 (41.0)	7 (17.9)	4 (10.3)	2 (5.1)	6 (15.4)	4 (10.3)
Peer Relationship Responses						
In the past 3 months how can you describe your experience regarding the relationship with peers	**Never n (%)**	**Sometimes n (%)**	**Regularly n (%)**	**Usually n (%)**	**Always n (%)**	**Uncertain n (%)**
1. Avoided talking, smiling, or laughing with other children because of your teeth, mouth, or face?	22 (56.4)	6 (15.4)	1 (2.6)	6 (15.4)	3 (7.7)	1 (2.6)
2.Been teased, bullied, or called names by other children because of your teeth, mouth, or face?	18 (46.2)	12 (30.8)	2 (5.1)	2 (5.1)	2 (5.1)	3 (7.7)
3. Been asked questions because of your teeth, mouth, or face?	7 (7.7)	13 (33.3)	4 (10.3)	1 (2.6)	12 (30.8)	2 (5.1)
4. Not wanted to meet new people because of your teeth, mouth, or face?	25 (64.1)	5 (12.8)	1 (2.6)	2 (5.1)	5 (12.8)	1 (2.6)
5. Been fighting or arguing with other children or family members because of your teeth, mouth, or face?	24 (61.5)	9 (23.1)	0 (0.0)	1 (2.6)	3 (7.7)	2 (5.1)
Family Impact Response						
In the past 3 months how can you describe the relationship of the child and the family	**Never n (%)**	**Sometimes n (%)**	**Regularly n (%)**	**Usually n (%)**	**Always n (%)**	**Uncertain n (%)**
1. Has the child condition cause any financial burden?	15 (38.5)	8 (20.5)	1 (2.6)	0 (0.0)	9 (23.1)	6 (15.4)
2. Have you or your partner/family member taken time out from work due to the child's condition?	12 (30.8)	12 (30.8)	1 (2.6)	2 (5.1)	8 (20.5)	4 (10.3)
3. Does your child require more attention from you?	9 (23.1)	10 (25.6)	2 (5.1)	1 (2.6)	11 (28.2)	6 (15.4)
4. Has the child's condition caused conflict in the Family internally	21 (53.8)	9 (23.1)	1 (2.6)	0 (0.0)	2 (5.1)	6 (15.4)

Oral Symptoms and Functional Wellbeing						
In the past 3 months have you had or experienced any of the following oral symptoms	**Never n (%)**	**Sometimes n (%)**	**Regularly n (%)**	**Usually n (%)**	**Always n (%)**	**Uncertain n (%)**
with siblings or other family members?						
5. Have you felt uncomfortable in public spaces or been ridiculed or stigmatized when with the child?	23 (59.0)	5 (12.8)	1 (2.6)	1 (2.6)	2 (5.1)	7 (17.9)
6. Have you or your partner/family member had concerns about the future wellbeing of the child?	12 (30.8)	10 (25.6)	1 (2.6)	1 (2.6)	4 (10.3)	11 (28.2)
7. Have you or the other parent felt upset because of the child's condition?	17 (43.6)	9 (23.1)	2 (5.1)	1 (2.6)	3 (7.7)	7 (17.9)
8. Have you or the other parent felt guilty for the child's state?	17 (43.7)	9 (23.1)	3 (7.7)	0 (0.0)	1 (2.6)	9 (23.1)
9. Has your sleep or that of your partner been affected?	22 (56.4)	6 (15.4)	2 (5.1)	0 (0.0)	2 (5.1)	7 (17.9)
General Health Response						
	Bad n (%)	Poor n (%)	Average n (%)	Good n (%)	Great n (%)	Uncertain n (%)
What is the general health of the child	0 (0.0)	0 (0.0.)	4 (10.3)	18 (46.2)	13 (33.3)	4 (10.3)

Appendix 9: Turnit-in Report

ORAL HEALTH-RELATED QUALITY OF LIFE IN CHILDREN WITH OROFACIAL CLEFTS WHO ARE TREATED AT A UNIVERSITY HOSPITAL

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