CURRICULUM VITAE

Steven Zinn, DMD
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EDUCATION

July 2022 – Present	**Residency in Orthodontics**- University of Maryland School of Dentistry -Certificate in Orthodontics expected June 2025 - Master of Science, May 2025 -Orthodontic skills: traditional braces, Damon system, custom brackets, clear aligner therapy, in-house aligners, surgical orthodontics, autotransplantation.
August 2018 - May 2022	**Dental School** (DMD)- **University of Pittsburgh School of Dental Medicine** -Class rank: 9/79
September 2014 - May 2018	**Bachelor of Science in Biology**- Lander College, a Division of Touro College (Queens, NY) - GPA: 4.0, Salutatorian

AWARDS, ACCOMPLISHMENTS, AND CERTIFICATIONS

May 2022	**Omicron Kappa Upsilon** (**OKU) -** inducted into national dental honor society
2019-2022	**Dean's List -** University of Pittsburgh School of Dental Medicine, earned all-academic years
Summer 2021	**Associate Level Teaching Certification** - Center for the Integration of Research, Teaching, and Learning-University of Pittsburgh School of Dental Medicine
2018	**Salutatorian -** Lander College, a Division of Touro College
2015-2018	**Dean's Honor List -** Touro College, earned all semesters

SCHOLARSHIPS

2021	The Roden Scholarship for Excellence in Prosthodontics Dr. Theodore B. Rosenthal Scholarship Sigmund Falk Scholarship
2020- 2021	Cheryl Cohen Scholarship Lyubor Nimax Scholarship Endowment Abe and Jean Comensky Memorial Scholarship
2020	AbbVie Immunity Scholarship

2019-2022	Dr. Lawrence S. Lipkind Endowment Stern Dental Scholarship
2015-2018	Touro Merit Scholarship
2014	HESC Achievement Award

PUBLICATIONS

Ickow IM, **Zinn S**, Stacy JM Jr, Martin B, Losee JE, D'Alesio A, Soxman J, Schuster LA. Dens Invaginatus in Patients with Cleft Lip and Palate: A Case Series. Cleft Palate Craniofac J. 2021 Mar 5:1055665621998534. doi: 10.1177/1055665621998534. Epub ahead of print. PMID: 33663245.

POSTERS

Zinn S, Diaz L, Mack J, Doughan M. Maintaining Maxillary Incisor Torque During Retraction with Clear Aligners. American Association of Orthodontists Annual Meeting. New Orleans, LA May 3-6, 2024.

Zinn S, Greene D, Diaz L, Sanchez D. Using Posterior Bite Plate Appliance to Reduce Anterior Open Bite Prior to Clear Aligner Treatment American Association of Orthodontists Annual Meeting. Chicago, IL April 21-24, 2023.

Diaz L, **Zinn S**, Mack, J, Pae EK. Asymmetric Extraction Considerations in Patients with an Overall Class I Malocclusion with a Unilateral Class II Molar Relationship Due to Missing Premolars. American Association of Orthodontists Annual Meeting. New Orleans, LA May 3-6, 2024.

Mack J, Diaz L, **Zinn S**, Ashton K, Bosio J. Orthodontic Camouflage of Class III Malocclusion in the Permanent Dentition Utilizing Reverse Pull Headgear Facemask, Maxillary Palatal Expansion, and Class III Elastics. American Association of Orthodontists Annual Meeting. New Orleans, LA May 3-6, 2024.

RESEARCH

June 2022- May 2025	**Master's Thesis, University of Maryland -** Oral Health Related Quality of Life in Patients with Cleidocranial Dysplasia -Performed oral examinations on over 100 individuals with CCD -Currently correlating exam findings with survey data to determine how oral manifestations of CCD affect quality of life
January 2020 - April 2020 *Pittsburgh, PA*	**University of Pittsburgh School of Dental Medicine – Center for Craniofacial Regeneration** -Researched amelogenin dephosphorylation and its effect on the regulation of toothenamel formation

June 2016 - August 2016 **Advanced Dermatology,**

Queens, NY - An Explanation for Adverse Reactions to Candida Albican Wart Injections

-Performed clinical research for a large dermatology practice.

TEACHING EXPERIENCE

August 2020 – May 2022 **Academic Career Track Area of Concentration (ACT ARCO)**
Pittsburgh, PA
-A two-year certification program to develop teaching and leadership skills for a career in academic dentistry.
-Taught first, second, and third-year dental students in didactic, pre-clinical, andclinical settings.

August 2019 - August 2020 **Peer Tutoring in Dental Education**
Pittsburgh, PA
-Courses tutored: Embryology, Oral Tissues, Microbiology, Cariology 1 (didactic and laboratory), Cariology 2 (didactic and laboratory), Physiology 1, Physiology 2, Gross Anatomy (didactic and laboratory), Pathology, and Immunology.

August 2017 - May 2018 **Peer Tutor at Lander College, a Division of Touro College**
Queens, NY
-Provided one-on-one tutoring in General Chemistry, Organic Chemistry, Biology and Biological Psychology.

June 14, 2021 **When the Appointment is No Longer About Dentistry**
Pittsburgh, PA
-Selected by school administration to present an hour-long lecture to the first-year dental class on how to manage microaggressions experienced in a dental practice.

Spring 2021- Spring 2022 **ADEA Academic Dental Careers Fellowship Program**
Pittsburgh, PA
-Participated in a one-year national fellowship which promotes teaching and research.

COMMUNITY SERVICE

September 2018 – May 2022 **Friendship Circle**
Pittsburgh, PA
-Volunteered weekly for an organization which provides supportand an inclusive community for individuals with special needs.

July 2015 - August 2018 **Chai Lifeline Hospital Volunteer**
New York, NY -Entertained children with cancer and other serious chronic illnesses in NewYork City hospitals and in their homes during weekly visits.

July 2015 - August 2018 **Chai Lifeline Big Brother**
Woodmere, NY -Visited a visually impaired teenager weekly in his home to engage him in social activities and facilitate peer interaction.

Summers 2015 - 2017 **Camp Simcha, a Project of Chai Lifeline**
Glen Spey, NY -Worked as a counselor in a sleepaway camp for children with cancer and otherchronic illnesses.

2019 - 2020 **Special Olympics Allegheny County**
Pittsburgh, PA -Coordinated sports activities for teens with special needs

LEADERSHIP AND ACTIVITIES

January 2020 – May 2022 **Founder and President of the Orthodontics Club,**
Pittsburgh, PA **University of Pittsburgh School of Dental Medicine**

June 2017 – August 2017 **Volunteer assistant, Five Towns Orthodontics**
Hewlett, NY

ACTIVE MEMBERSHIPS

American Dental Association (ADA), American Association of Orthodontists (AAO), Middle Atlantic Society of Orthodontics (MASO), Maryland State Society of Orthodontics (MSSO), Maryland State Dental Association (MSDA)

ADDITIONAL CERTICFICATIONS

Maryland State Dental License
CDCA-WREB
ABO Written Exam. Oral exam to be taken after residency is completed.

ABSTRACT

Title: Oral Health-Related Quality of Life in Patients with Cleidocranial Dysplasia

Steven Zinn, Master of Biomedical Sciences, 2025

Thesis Directed by: Monica Schneider, DDS, MS, Clinical Associate Professor

Background: There are many dental manifestations of cleidocranial dysplasia (CCD). These include retained primary teeth, supernumerary teeth, and impacted permanent teeth.[1] Additionally, these individuals may also have anterior crossbites, posterior crossbites, and reduced overbite.[23] Patients with CCD often require extensive orthodontic treatment to erupt the impacted permanent dentition and often also require orthognathic surgery to correct the malocclusion. Additionally, some patients with CCD undergo prosthodontic replacement of impacted teeth rather than orthodontics, or a combination of prosthodontic and orthodontic treatment. This study focuses on whether individuals with CCD with certain malocclusion traits have a worse oral health related quality of life (OHQOL) than those without these malocclusion traits. Additionally, this study aims to determine if individuals with CCD that received orthodontic treatment with or without prosthodontics had a better OHQOL than those treated with prosthodontics alone.

Methods: Individuals over the age of 15 with CCD were recruited to undergo an oral examination to assess malocclusion and number of missing teeth and to complete the OHIP-14 survey. This survey considered the following domains: Functional Limitation, Physical Pain, Psychological Discomfort, Physical Disability, Psychological Disability, Social Disability, and Handicap. Independent sample t-tests were performed to compare OHIP-14 scores in individuals with CCD with and without specific oral findings.

Additionally, individuals with CCD who had completed their orthodontic treatment were asked about the duration of their orthodontic treatment, and if in hindsight, they would have preferred prosthodontic treatment alone.

Results: Seventy-five individuals with CCD over the age of 15 underwent oral exams, and 61 of these individuals completed the OHIP-14 survey. Independent sample t-tests revealed that individuals with CCD who had posterior crossbites experienced significantly worse oral health-related quality of life (OHQOL) in the domain of physical pain ($p = 0.015$, 95% CI: [0.316, 2.750]). For those with anterior open bites, a significantly worse OHIP-14 score was observed in the domain of handicap ($p = 0.046$, 95% CI: [0.027, 2.919]). Anterior crossbites resulted in statistically significant worse OHIP-14 score in the functional limitation domain ($p = 0.027$, 95% CI: [0.149, 2.373]). Lastly, individuals who received only prosthodontic treatment and no orthodontic treatment reported significantly worse overall total OHIP-14 scores ($p = 0.037$, 95% CI: [0.685, 21.015]) and higher scores in physical pain ($p = 0.038$, 95% CI: [0.114, 3.981]), physical disability ($p = 0.005$, 95% CI: [0.872, 4.543]), and social disability ($p = 0.020$, 95% CI: [0.091, 3.398]). Nineteen individuals who completed their orthodontic treatment responded to the survey about their orthodontic treatment. Orthodontic treatment among these respondents took an average of almost ten years, and 40% of these respondents stated that they would have preferred prosthodontic treatment alone.

Conclusion: Individuals with CCD who have anterior crossbites, posterior crossbites, or anterior open bites have statistically significantly worse OHQOL in specific domains compared to individuals with CCD without these malocclusions. Additionally, although 40% of individuals with CCD who had orthodontic treatment stated that they would have

preferred prosthodontics alone, OHIP-14 results indicate that those who had received prosthodontics alone had statistically significant worse OHQOL.

Oral Health Related Quality of Life in Patients with Cleidocranial Dysplasia

by
Steven Zinn

Thesis submitted to the Faculty of the Graduate School of the
University of Maryland, Baltimore in partial fulfillment
of the requirements for the degree of
Master of Science
2025

ACKNOWLEDGMENTS

I would like to express my heartfelt gratitude to everyone who contributed to making this research possible.

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LIST OF ABBREVIATIONS

CCD	Cleidocranial Dysplasia
OHQOL	Oral Health Related Quality of Life
OHIP	Oral Health Impact Profile

1. Introduction

1.1 Cleidocranial Dysplasia: Its Features and Genetic Basis

Cleidocranial Dysplasia (CCD) is a rare autosomal dominant genetic disorder which affects one in one million individuals.[1,2] Classic features of CCD, a disorder which affects teeth and bones, include missing or small clavicles, open fontanelles, over-retained primary teeth, supernumerary teeth, and impaired eruption of permanent teeth. Additionally, individuals with CCD may have short stature, hearing loss, an obstructed airway, and other bone deformities.[2,3]

A diagnosis of CCD is made based on clinical and radiographic findings or from genetic testing which reveals a mutation in the *RUNX2* gene.[3,4] CCD has an autosomal dominant mode of inheritance, and there is a high incidence of de novo mutations which cause this disorder.[3] About 70% of individuals with CCD have a mutant variant in *RUNX2*, while 30% of individuals with CCD have no known genetic mutation.[5] Additionally, the variable phenotype and genotype are not entirely understood.

RUNX2 is fundamental for proper skeletal development. This transcription factor plays a crucial role in the differentiation of osteoblasts from multipotent stem cells, chondrocyte development, determination of phenotype of cartilage mesenchymal cells, and bone formation.[6] A reduction in osteoblasts and osteoblastic activity caused by a mutation in *RUNX2* can negatively affect osteoclast activation.[7] Osteoblasts carry the membrane-bound protein RANKL (receptor activated nuclear factor kappa B ligand). Osteoclasts have RANKL receptors which are activated in the presence of RANKL. In the absence of

osteoblasts, the paucity of RANKL may lead to reduced osteoclastic activity. Osteoclasts are vital to bone resorption necessary for dental eruption. If osteoclastic activation is reduced, dental eruption will be significantly delayed.[7] In 2004, Yoda et al performed an experiment in which the dental eruption of heterozygous *RUNX2* knockout mice was compared to wild type mice. The authors observed, as predicted, that in the knockout mice, both maxillary and mandibular dental eruption was considerably delayed. Additionally, although both types of mice had an increase in alveolar bone resorption and osteoclastic recruitment and activity in postnatal days 8 to 10, in mutant mice, this increase was significantly reduced. These findings suggest that deficient recruitment of osteoclasts caused by a mutation in *RUNX2* may be one of the cellular mechanisms that cause delayed eruption in individuals with CCD.[4]

1.2 Medical and Dental Care for Individuals with CCD

Individuals with CCD should be evaluated and treated by a multidisciplinary team. Specifically, oral surgeons, orthodontists, and prosthodontists aid in the eruption of permanent teeth and restoration of missing or damaged teeth. Pediatric dentists and general dentists ensure the health of the dentition, which is especially significant given the prolonged maintenance of primary teeth and lengthy duration of orthodontic treatment. Additionally, maxillofacial surgeons may be required for future orthognathic surgery when the patient's growth is complete.[8] Otolaryngologists evaluate patients with CCD for hearing loss and obstructive sleep apnea, and endocrinologists assess bone density for osteoporosis.[3] Geneticists and primary care physicians aid in diagnosis of CCD and

coordination of care, and physiatrists or physical therapists treat patients with CCD who have skeletal pain and malfunction.

Dental eruption consists of two phenomena: resorption of bone and overlying primary tooth roots, and a propulsive mechanism for the permanent tooth.[9] If either of these stages of eruption does not occur, a tooth will not emerge or come into occlusion. Failure of eruption of permanent teeth in individuals with CCD is due to a lack of resorption of alveolar bone and primary teeth, supernumerary teeth, or heavy overlying fibrous gingiva which obstructs the eruption pathway.[9]

To aid in the eruption of the impacted permanent dentition, surgery is performed to extract the primary and supernumerary teeth and expose the permanent teeth which are to be maintained. Exposing permanent teeth includes creating a flap or window in the gingiva overlying the impacted tooth, removing alveolar bone which covers the dental crown, and bonding a chain or bracket to the exposed tooth. After exposure, orthodontists apply traction to bring each impacted tooth into proper occlusion.

1.3 Orthodontic Techniques to Treat Patients with CCD

Patients with CCD are among the most difficult to treat orthodontically. Previously, before orthodontics was seen as a viable treatment option in these individuals, primary teeth were extracted and the unerupted permanent teeth were replaced with a prosthesis.[10] Currently, there are four main orthodontic treatment protocols for CCD: the Jerusalem method, the Toronto-Melbourne method, the Belfast-Hamburg method, and the Bronx method.

The Jerusalem approach was described by Dr. Adrian Becker in four separate editorials which comprise four distinct phases of orthodontic treatment for individuals with CCD.[11-14] In his papers describing this approach, Dr. Becker discusses that the dental age of those with CCD is three years behind that of their peers. Orthodontic exposures should not be performed until half to two thirds of the tooth root has formed. Hence, while most children would be ready for impacted incisor exposure at the age of seven, individuals with CCD are not ready for this treatment until the age of nine or ten. Additionally, patients with CCD often have skeletal class III malocclusions with anterior crossbites which is usually due to midface hypoplasia. While waiting for proper permanent incisor root development, class III correction with facemask can be performed.[11] Correction of the skeletal class III represents the first phase of the Jerusalem Method.

The second and third phases of the Jerusalem method entail bringing the permanent teeth into occlusion.[12] In individuals with CCD, the first molars usually erupt autonomously, and one or more incisors may erupt partially as well. However, often, the first molars are only teeth to erupt spontaneously. In this second phase of the Jerusalem Method, the permanent maxillary and mandibular incisors (and at times, the mandibular canines) are exposed and brought into the dental arch when half to two third root formation is seen radiographically. At the age of 10, the incisors usually meet this criterion. Oral surgery is performed to extract the primary teeth and supernumerary teeth in the anterior region. The first molars and posterior deciduous teeth are used as anchorage to erupt the exposed permanent incisors. The posterior deciduous teeth are not extracted at this time in order to maintain some posterior teeth (other than first permanent molars) until the succedaneous posterior teeth can be exposed and brought into occlusion three to four years

later.[12] Around the age of 13-14, when there is adequate root formation in the permanent canine and premolar teeth, the premolar phase of tooth eruption (phase III) occurs. In this phase, the remaining deciduous and supernumerary teeth are removed, the permanent canines and premolars are exposed, and force is applied to bring them into occlusion.[13]

The final phase of orthodontic treatment in patients with CCD is preparing for orthognathic surgery if indicated.[14] Although one can pause between phases of orthodontic treatment, in many cases, when one phase is complete, the next phase is ready to begin.

In the Toronto-Melbourne protocol, treatment is initiated at the age of five or six. At that time, all the anterior deciduous and supernumerary teeth are removed with the hope of autonomous eruption of permanent teeth.[15] Since the major contributor to delayed eruption in individuals with CCD is lack of overlying bone resorption, it is possible that removing the primary teeth, supernumerary teeth, and bone overlying the permanent teeth will enable spontaneous eruption.[8] In this method, teeth are only exposed if they remain unerupted. The same process is performed on the posterior teeth at the age of 9 to 10.[15] According to a 2018 study, removing the primary and supernumerary teeth alone enabled the permanent maxillary and mandibular anterior teeth to spontaneously erupt approximately 55% of the time, the maxillary canines and premolars to erupt about 40% of the time, and the mandibular canines and premolars to erupt less than 30% of the time.[16]

The Belfast-Hamburgh method includes only one surgery in which all the primary and supernumerary teeth are removed simultaneously, and permanent teeth are exposed and bonded to bring them into proper occlusion.[14] This method reduces the number of surgical appointments to one invasive surgery.[15]

The Bronx method is similar to the Belfast-Hamburgh method, however, while waiting for the permanent teeth to be brought into occlusion, the patient is given a removable partial denture for functional and esthetic purposes.[15]

Orthodontic treatment time in individuals with CCD may take ten or more years.[1] This lengthy orthodontic treatment puts these patients at risk for caries, increased root resorption, and treatment burnout. In the most unfortunate circumstances, after many years of orthodontic treatment, patients may need extensive prosthodontic work to replace carious teeth and teeth with inadequate periodontal coverage after exposure and forced eruption.

Due to the lengthy orthodontic treatment time as well as the number of surgeries needed in individuals with CCD, patients may elect to pursue prosthodontics as a treatment modality rather than orthodontics. These treatments can include removing all impacted teeth and placing implants to restore the missing dentition, fabricating dentures, or placing an implant-supported denture.[17,18]

1.4 Oral Health Related Quality of Life in Individuals with CCD

It is not known how the dental manifestations of CCD affect the oral health related quality of life (OHQOL) of individuals with CCD. There have been many studies regarding other syndromes and oral health related quality of life. For example, a study was done on individuals with Down syndrome and their families which concluded that 34.9% of individuals with Down syndrome and 46% of family members have a quality of life which

is negatively affected by oral health. The reduced OHQOL was mainly due to dental pain and the family's emotional response to this pain.[19] Similar studies have been performed on individuals with Ehlers Danlos and Burning Mouth syndromes.[20,21] In a cross-sectional study titled "Malocclusion Traits and Oral Health Related Quality of Life in Children with Osteogenesis Imperfecta," Najirad et al. correlated findings from oral examinations with results from validated OHQOL surveys. The authors determined which specific malocclusion traits led to reduced oral health-related quality of life in specific domains. The study concluded that adolescents with osteogenesis imperfecta who have posterior open bites or crossbites have a reduced OHQOL due to functional limitations and oral symptoms.[22] Similarly, it is presumable that individuals with CCD who have many dental abnormalities may also have a reduced OHQOL.

1.5 Previous Publications about Cleidocranial Dysplasia

There have been many publications about the dental and craniofacial findings of CCD. In addition to retained deciduous teeth and impacted supernumerary and permanent teeth, individuals with CCD have increased incidence of anterior and posterior crossbites and reduced overbite.[23] However, because CCD is so rare, the sample sizes of these studies which report on malocclusions in CCD are usually quite small. For example, a study on cephalometric findings in those with CCD, which concluded that individuals with CCD had shorter anterior cranial bases and hypodivergent mandibles, included 5 individuals with CCD.[24] Interestingly, this study noted that although individuals with CCD have hypoplastic maxillae, the SNA measurements of these individuals are greater than those

who are unaffected due to their short anterior cranial base measurements. A longitudinal study published in 1981, which concluded that individuals with CCD have short maxillary heights due to reduced resorption of the nasal floor and reduced apposition on the orbital floor, examined 22 affected individuals.[25] Even systematic reviews have difficulties reaching triple digit sample sizes when researching rare conditions such as CCD. For instance, a systematic review regarding the demographic, clinical, and radiological characteristics of CCD in the entire South America garnered a sample size of 72.[26] This study documented that 46.5% of the affected individuals were brachycephalic and 43.3% had maxillary hypoplasia. A 2018 systematic review[23] included 60 publications which contained 79 patients with CCD and noted that among both males and females, 100% had maxillary transverse constriction, supernumerary teeth, and eruption failure, 94% had maxillary hypoplasia, and six individuals received orthognathic surgery to correct their malocclusion. Additionally, 98% of individuals had a class III dental relationship.[23] One review study regarding the dentomaxillofacial variability seen in CCD had a sample size of 283 patients. However, this study included 40 single case presentations and 17 multiple case studies to reach this high sample size.[27]

1.6 CCD Smiles

CCD Smiles is the world's largest patient support group for CCD. This organization hosts a biennial conference for patients with CCD and multiple virtual support groups on social media, including patient and caregiver Facebook support groups. The annual

meetings, aside from being a source of education, inspiration, and camaraderie for people with the same rare syndrome, are excellent opportunities for research.

The Johns Hopkins School of Medicine collaborated with CCD Smiles at two such meetings. The primary goals of the Johns Hopkins research are to understand the range of physical characteristics of CCD through physical examinations, examine the genetic etiology of CCD by collecting saliva samples and obtaining pedigree data, and link the genetic data to physical features. Moreover, the study aims to investigate the quality of life and oral health related quality of life of patients with CCD, and to identify the diverse needs of these patients. Another goal is to create standardized guidelines for diagnosis and management of CCD. This project was a collaborative effort between the Department of Plastic and Reconstructive Surgery and the Department of Genetic Medicine.

1.7 Research Objectives

The purpose of this study is to describe the impact of dental manifestations of CCD on Oral Health-Related Quality of Life (OHQOL) in individuals aged 15 and above.

Research Question 1:

In individuals with CCD, does the presence of an anterior crossbite, posterior crossbite, anterior open bite, or eight or more missing teeth lead to a different Oral Health-Related Quality of Life (OHQOL) than the OHQOL seen in individuals with CCD who do not have these oral findings?

Null Hypothesis 1(H_0):

There is no significant difference in the OHQOL between individuals with CCD that have an anterior crossbite, posterior crossbite, anterior open bite, or eight or more missing teeth, and those with CCD without these findings.

Alternative Hypothesis 1(Ha):

There is a significant difference in the OHQOL between individuals with CCD that have an anterior crossbite, posterior crossbite, anterior open bite, or eight or more missing teeth, and those with CCD without these findings.

Research Question 2:

In individuals with CCD, do those who underwent prosthodontic treatment without orthodontic treatment have different OHQOL than the OHQOL seen in individuals with CCD who underwent orthodontic treatment with or without prosthodontic treatment?

Null Hypothesis 2(H_0):

There is no significant difference in the OHQOL between individuals with CCD who underwent prosthodontic treatment without orthodontic treatment, and those who underwent orthodontic treatment with or without prosthodontic treatment.

Alternative Hypothesis 2(Ha):

There is a significant difference in OHQOL between individuals with CCD who underwent prosthodontic treatment without orthodontic treatment, and those who underwent orthodontic treatment with or without prosthodontic treatment.

Variables:

In this study, the independent variables include the type of dental treatment and the presence of intraoral features (anterior crossbite, posterior crossbite, anterior open bite, or eight or more missing teeth), while the dependent variable is the OHQOL of individuals with CCD.

2. Materials and Methods

IRB approval was obtained from Johns Hopkins School of Medicine (IRB00246592). The University of Maryland, Baltimore formed a reliance agreement with Johns Hopkins School of Medicine to allow for institutional collaboration.

2.1 Sample

Study participants were recruited at two conferences organized by CCD Smiles: one held in Salt Lake City, Utah, in June 2022, and another in London, England, in November 2022. One hundred thirty one individuals with CCD attended the meeting held in Salt Lake City and 72 individuals with CCD attended the meeting in London. Inclusion criteria required participants to be members of CCD Smiles, have a confirmed diagnosis of CCD, be at least 15 years old, and able to complete a survey in English independently. Exclusion criteria included individuals under 15 years of age, those unable to complete the survey in English without assistance, and anyone without a diagnosis of CCD.

2.2 Data Collection

At both conferences, written consent was obtained from participants after informing them of the study procedures and protocols. After consent was obtained, participants were asked to undergo an oral examination and complete a validated survey.

The oral examinations were performed by four individuals: two craniofacial orthodontists, one craniofacial orthodontic fellow, and one orthodontic resident. The examiners were calibrated to ensure that their definitions of dental findings were consistent. The information collected at the oral exam and recorded on paper included demographic information and extraoral and intraoral characteristics. Demographic data obtained included the patient's birthdate, sex, and unique study number. Extraoral characteristics included whether the facial profile was concave, convex, or straight, whether the individual was dolichofacial, brachyfacial, or mesofacial, and whether the examinee had a facial asymmetry.

After completing the extraoral examination, the clinicians evaluated the participants intraorally. The examinees' palates were examined and classified as being normal, high arched, or clefted, the teeth were counted to see how many were missing, and their Angle's molar classification was noted. Additionally, the individual's overbite, overjet, and degree of crowding or spacing was calculated and the presence of an anterior crossbite, unilateral posterior crossbite, or bilateral posterior crossbite, was noted. Lastly, the oral examinator noted whether the patient had braces on, had any prosthetic teeth, or had a history of tooth exposure during orthodontic treatment (See Oral Exam Form, Appendix A).

After the oral examination, examinees completed the oral health quality of life survey. Surveys were either taken on personal devices by scanning a QR code, or on an iPad which was provided. These qualified surveys were created using Qualtrics, taken unassisted, and were age specific. Children ages 7-10 took the CPQ 8-10 qualified survey[28], adolescents ages 11-14 took the CPQ 11-14 survey[29], and those over 14 took the OHIP-14

(Oral Health Impact Profile) survey.[30] The three surveys differed in the number of questions asked and the age-appropriate language of the questions. This portion of the study focuses on participants ages 15 and above and the OHIP-14 survey.

2.3 OHIP-14 Survey

The OHIP-14 survey has 14 questions regarding 7 different domains (Appendix B). These domains are: Functional Limitation, Physical Pain, Psychological Discomfort, Physical Disability, Psychological Disability, Social Disability, and Handicap. Two questions were asked per domain. Each question asked about how the oral condition affected the individual in the past 12 months. The answer choices to each of the fourteen questions were based on frequency of the events: never, 0; hardly ever, 1; occasionally, 2; fairly often, 3; and very often, 4. The answers to the questions in each domain were summed, and the answers to all the questions were summed to quantify the total OHIP-14 score. The highest score an individual can obtain per domain was 8 and the highest possible score for the entire survey was 56. The greater the value of the score, the worse the oral health related quality of life. Also, the survey asked for the same demographic data collected during the oral exam to enable correlation between the oral exam findings and the results from the OHIP-14 survey.

Furthermore, participants older than 18 were asked in an additional survey if they completed orthodontic treatment, and if so, how many years it took to complete. These individuals were also asked if in hindsight they would have preferred to receive

prosthodontic treatment alone instead of orthodontics or a combination of orthodontics and prosthodontic treatment.

2.4 Oral Exam Findings to be Compared with OHIP-14 Results

This study focused on comparing four specific oral exam findings with each of the seven domains from the OHQOL survey. These exam findings included: number of missing teeth, presence of an anterior crossbite, presence of a posterior crossbite, and presence of an anterior open bite. Additionally, the OHQOL of those who received prosthodontics alone was compared to those who received either orthodontics alone or a combination of prosthodontics and orthodontics.

The count of missing teeth excluded those replaced by fixed prostheses, such as bridges or implants. Examinees with fewer than eight missing teeth were assigned a value of 0, while those missing eight or more teeth (excluding third molars) were assigned a value of 1. Assigning categories to numerical values facilitates statistical analysis. The threshold of eight missing teeth was chosen based on evidence suggesting that "the prevalence of negative impacts increases sharply once the number of teeth present drops below 20."[32]

Anterior crossbite was considered present when any of the six maxillary anterior teeth were lingual to their mandibular counterparts. The presence or absence of anterior crossbite was recorded as 0 equals no and 1 equals yes.

Posterior crossbite was considered present when any of the maxillary posterior teeth were lingual to their mandibular counterparts. The presence or absence of posterior crossbite was recorded as 0 equals no and 1 equals yes.

Anterior open bite was considered present when there was a lack of overlap of the maxillary and mandibular incisors which was not due to normal eruption. The presence or absence of anterior open bite was recorded as 0 equals no and 1 equals yes.

An examinee with CCD was considered to have received prosthodontics when they had a mode of replacement teeth such as implants, bridges, or fixed or removable complete or partial dentures. If an individual had dental crowns, they were not classified as having received prosthodontics since crowns do not replace missing teeth. These individuals were considered to have had orthodontics if they had a history of exposures, braces, or cross arch space maintainers such as lower lingual holding arches, trans palatal arches, or Nance appliances. If the examinee had a history of orthodontics or orthodontics and prosthodontics, they were assigned the number 0 and if they received prosthodontics alone, they were assigned to the number 1.

2.5 Statistical Analysis

Independent sample t-tests were performed to examine the impact of oral findings on the Oral Health Impact Profile (OHIP-14) scores and its subdomains, comparing individuals with CCD with and without specific oral findings. The significance level for this analysis was set at 5%. Levene's test was used to test the assumption of equal variances

being compared and Cohen's d was used to measure the effect size and magnitude of the differences between the OHIP-14 scores of those with malocclusions and those without.

3. Results

A total of 104 individuals with Cleidocranial Dysplasia had oral examinations at the CCD Smiles meetings. There were 79 individuals who participated in the research at the June 2022 meeting in Salt Lake City, and there were 25 individuals who participated in the research at the November 2022 meeting in London. Of these 104 individuals, 75 were aged 15 or older. Individuals under age 15 were excluded since this research focuses on individuals 15 or older.

3.1 Oral Examinations

Of the 75 individuals 15 years of age and older who had oral examinations, 70% had brachyfacial profiles and 80% had high arched palates. Forty-four percent of those examined had concave profiles, 23% had anterior crossbites, and 47% had a bilateral Angle's class III molar relationship or a Class III subdivision molar relationship in which one set of molars had an Angle's Class III relationship, and the contralateral set of molars had an Angle's class I relationship. Forty-nine percent of the examinees had unilateral or bilateral posterior crossbite, 11% had anterior open bites, and 32% had eight or more missing teeth.

Most patients received orthodontics alone (57%) to treat the impacted teeth, while 19% of patients received orthodontics and prosthodontics, 12% had prosthodontics alone, and 12% received neither orthodontics nor prosthodontics. (See Table 1 for demographic and Figures 1-12 for additional intraoral and extraoral findings.)

Demographic Data

Participants aged 15+, No.	75
Female, No(%)	48 (64)
Male, No(%)	27 (36)
Age, y, Mean (SD)	35.4 (14.4)
Age, y, Mean (SD) Female	35.8 (12.6)
Age, y, Mean (SD) Male	34.7 (17.4)

Table 1: Demographic Data of Individuals with CCD over the age of 15 who completed an oral examination

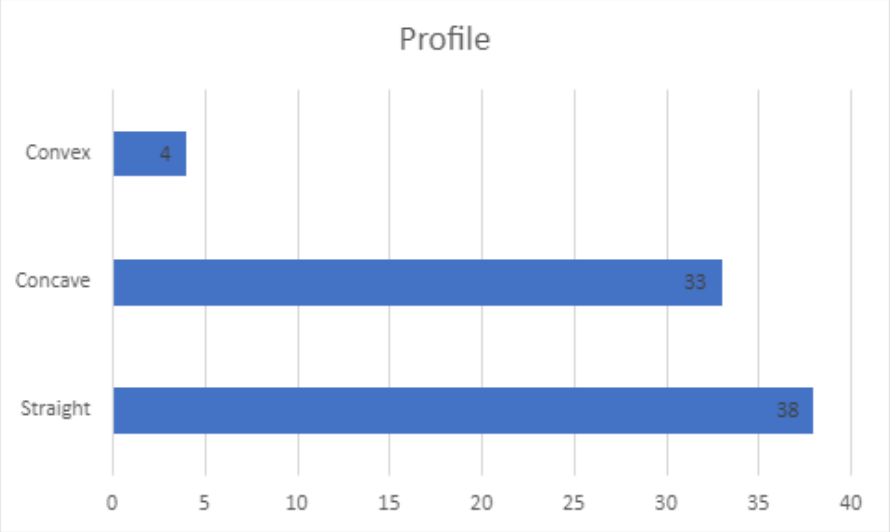


Figure 1: Profile type of individuals with CCD over the age of 15 who completed an oral examination. Determined by visualizing a line from glabella to subnasale and from subnasale to menton in lateral view.

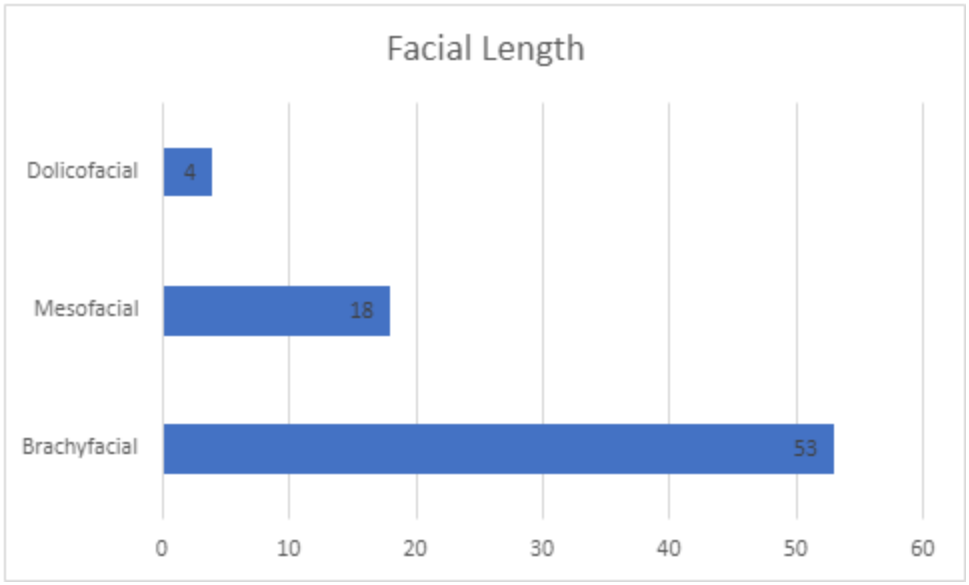


Figure 2: Facial length of individuals with CCD over the age of 15 who completed an oral examination.

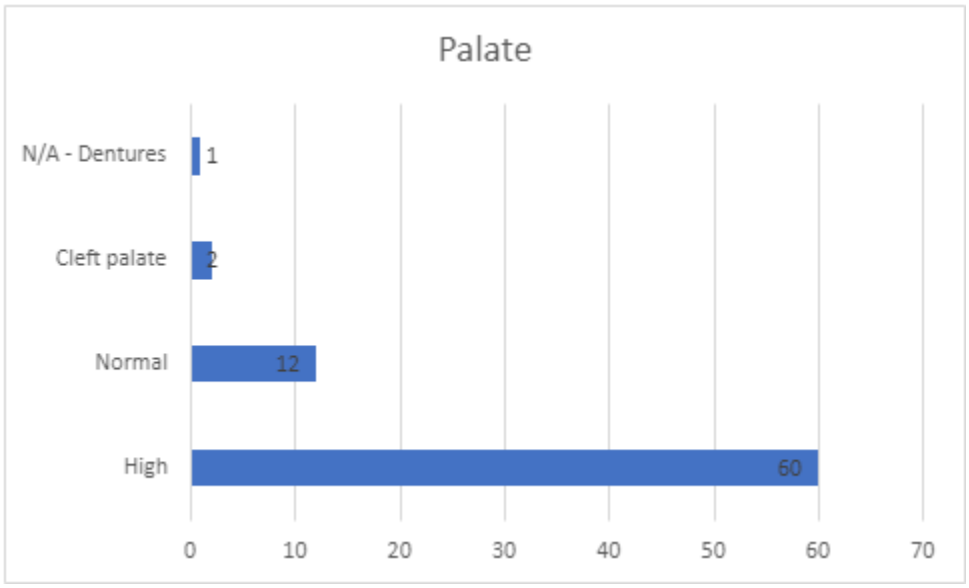


Figure 3: Classification of palate depth in individuals with CCD over the age of 15 who completed an oral examination. One patient did not want to remove dentures, and a classification could not be performed.

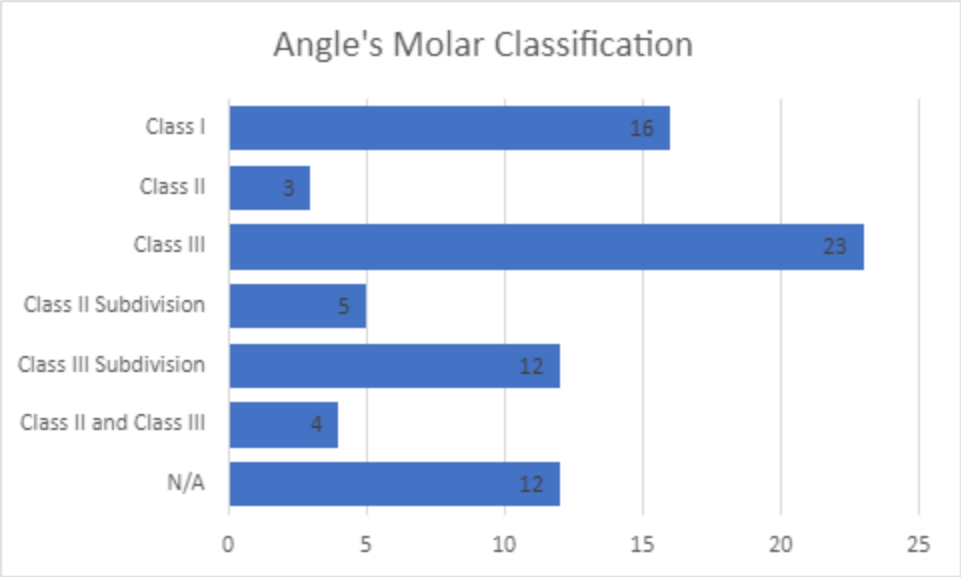


Figure 4: Angle's molar classification in individuals with CCD over the age of 15 who completed an oral examination. A half cusp class II or end on class II was classified as "Class II". If the first molars were missing, no classification was given.

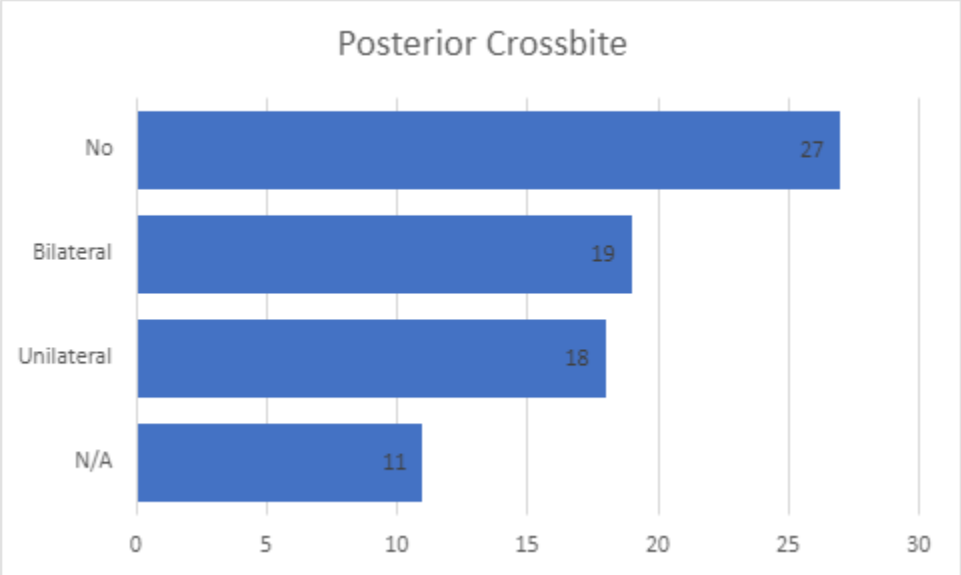


Figure 5: Presence of posterior crossbite in individuals with CCD over the age of 15 who completed an oral examination. If all posterior teeth were missing, no classification was given.

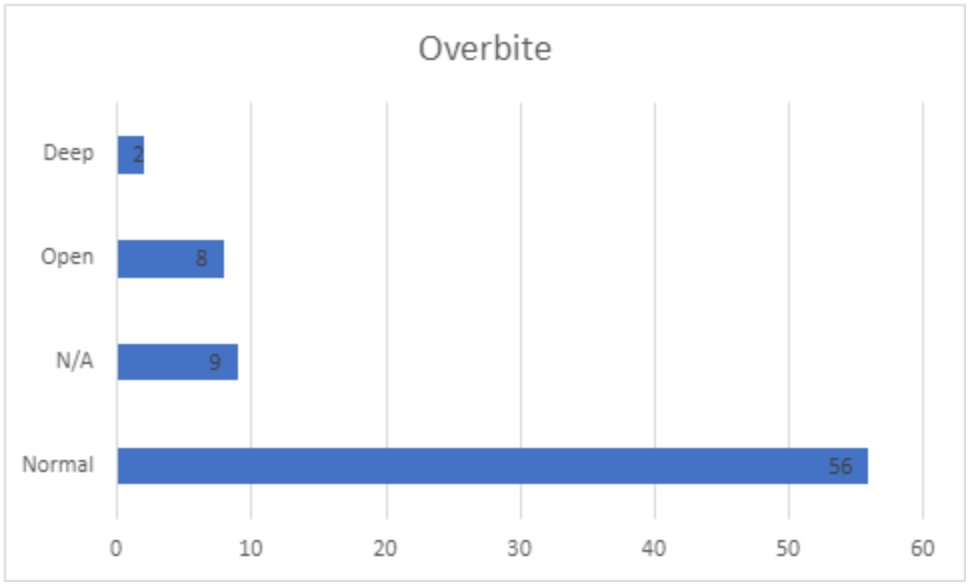


Figure 6: Overbite classification in individuals with CCD over the age of 15 who completed an oral examination. Overbite 0-4mm was considered normal, 5+mm was considered deep and a negative overbite was considered open. If all incisors were missing, no classification was given.

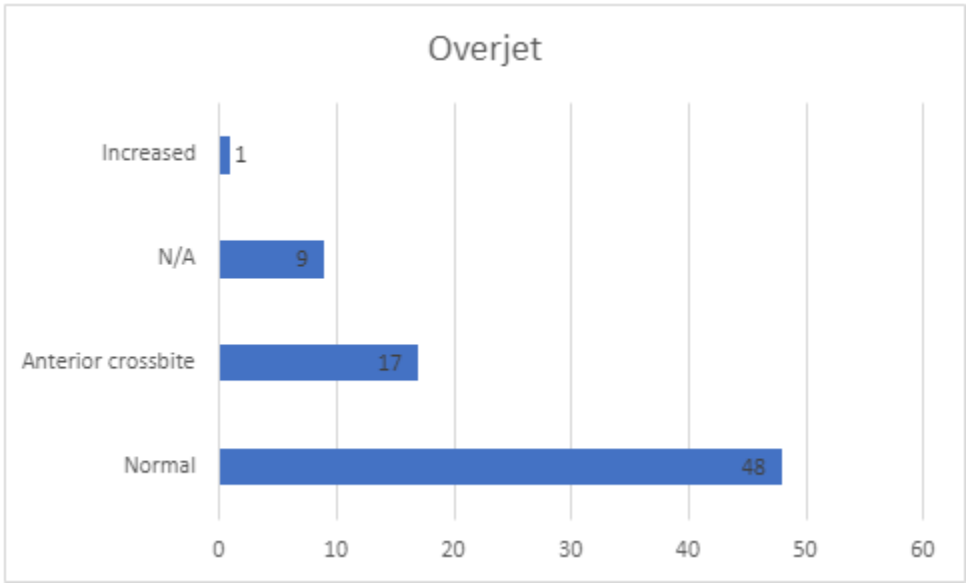


Figure 7: Overjet classification in individuals with CCD over the age of 15 who completed an oral examination. Overjet 0-4mm was considered normal, 5+mm was considered increased, and a negative overjet was classified as an anterior crossbite. If all incisors were missing, no classification was given.

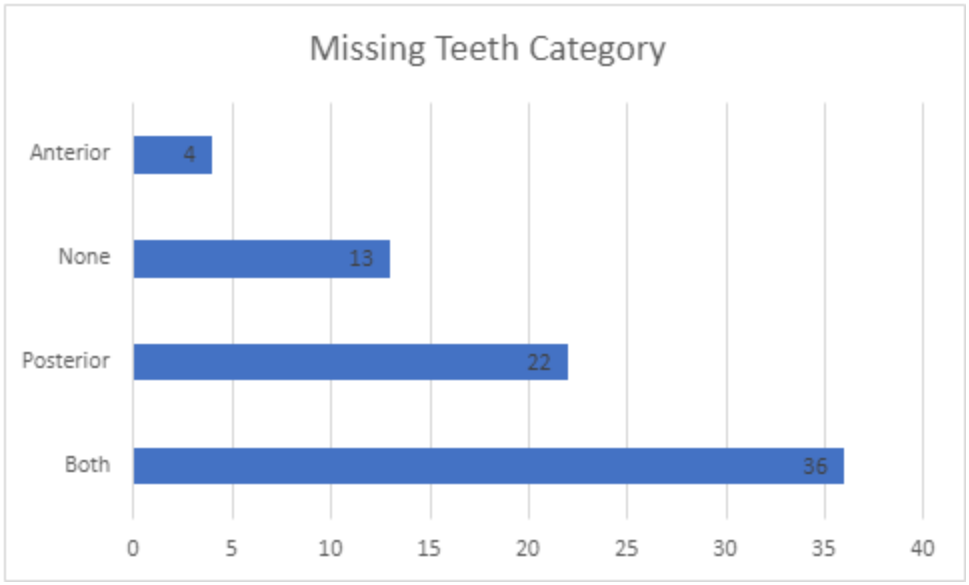


Figure 8: Missing tooth category in individuals with CCD over the age of 15 who completed an oral examination.

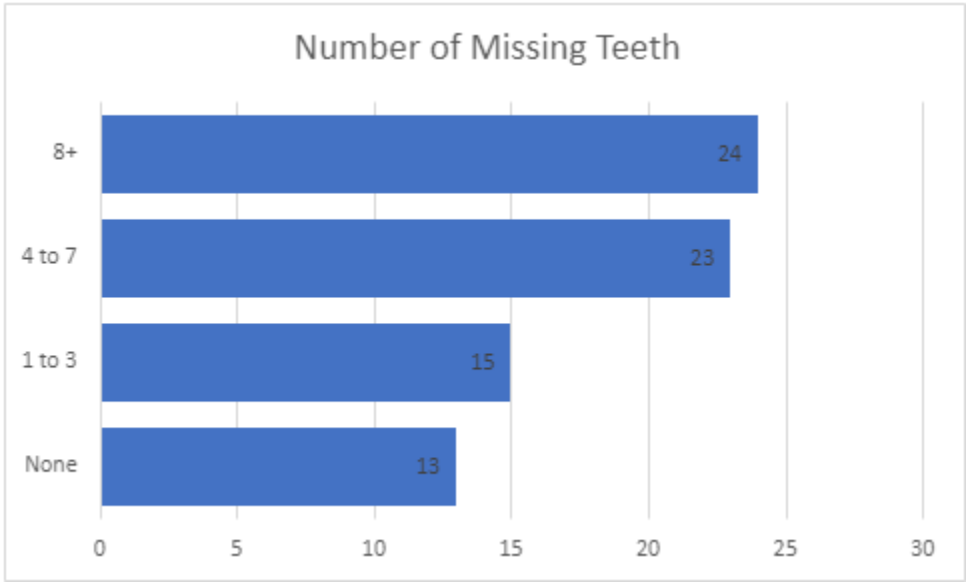


Figure 9: Number of missing teeth categorized in individuals with CCD over the age of 15 who completed an oral examination.

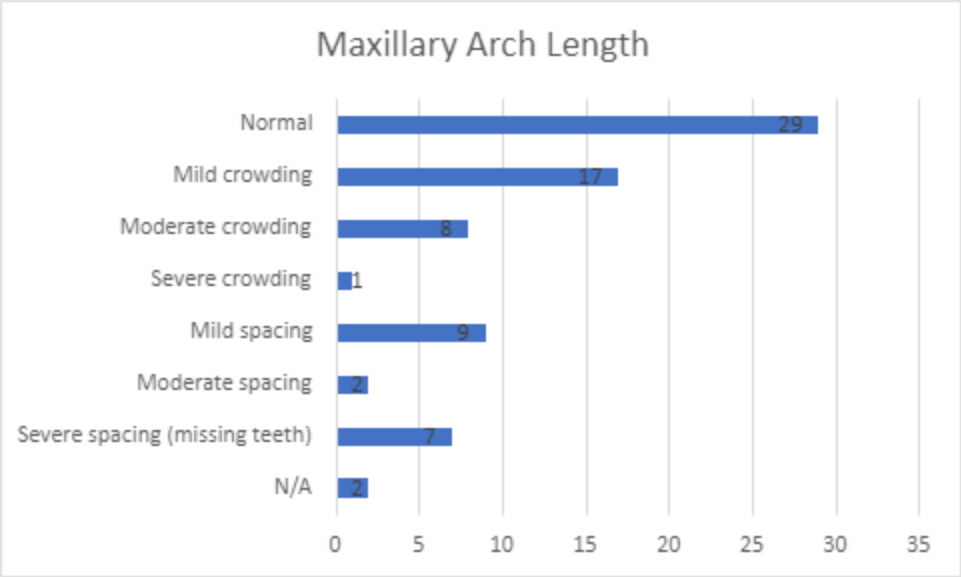


Figure 10: Classification of crowding or spacing in maxilla in individuals with CCD over the age of 15 who completed an oral examination. Normal: no crowding or spacing, Mild: 0-4mm of crowding or spacing, Moderate: 4-8mm of crowding or spacing, Severe: more than 8mm of crowding or spacing.

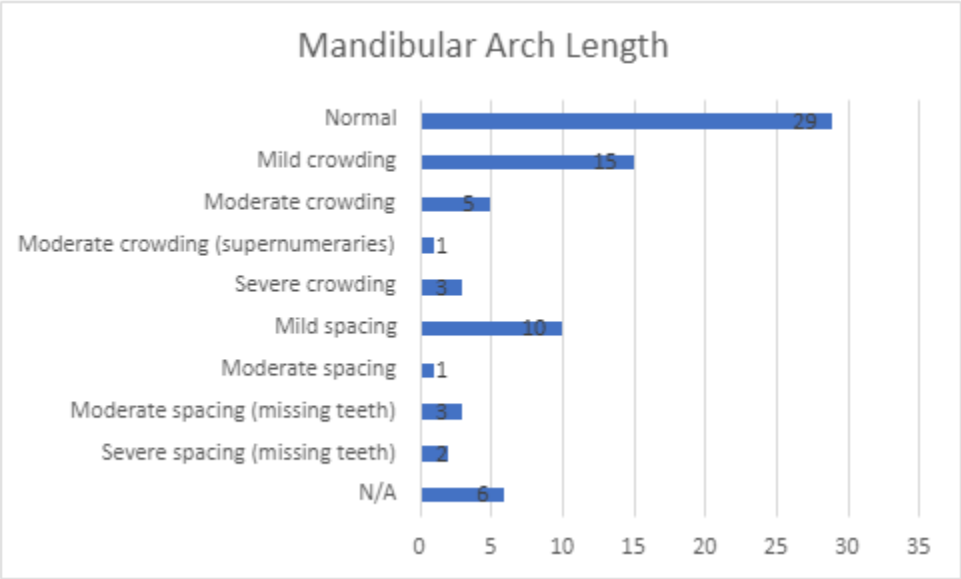


Figure 11: Classification of crowding or spacing in mandible in individuals with CCD over the age of 15 who completed an oral examination. Normal: no crowding or spacing, Mild- 0-4mm of crowding or spacing, Moderate: 4-8mm of crowding or spacing, Severe: more than 8mm of crowding or spacing.

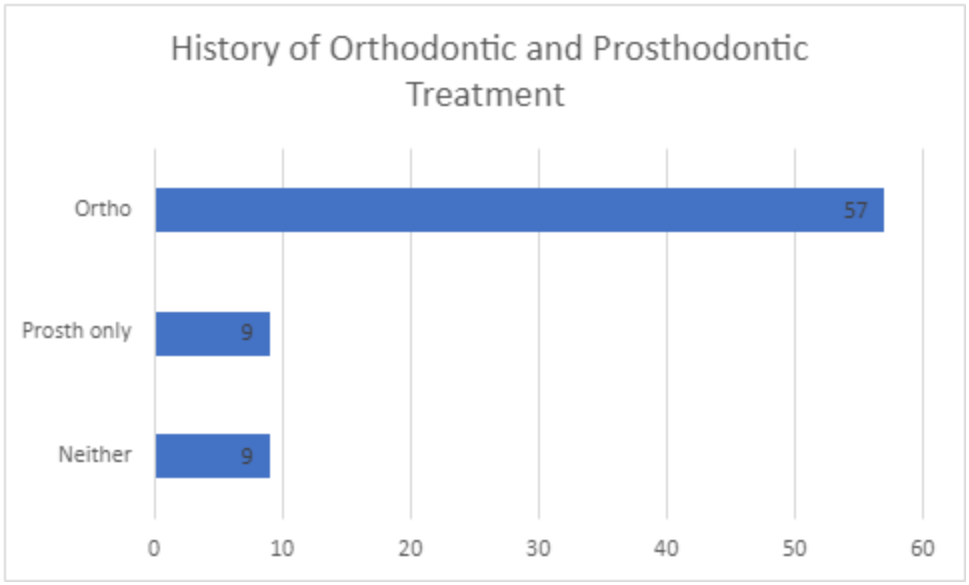


Figure 12: History of orthodontic and prosthodontic treatment in individuals with CCD over the age of 15 who completed an oral examination. Category title "Ortho" includes individuals who had both orthodontic treatment alone or orthodontic treatment and prosthodontic treatment.

3.2 Oral Examination and OHIP-14

Of the 75 individuals with CCD who underwent oral examinations, 61 completed the OHIP-14 qualified survey. The oral examination findings of these 61 individuals which were compared with the OHIP-14 survey results (posterior crossbite, anterior open bite, anterior crossbite, missing 8+ teeth, and history of prosthodontics without orthodontics) are seen in Table 2. If participants did not have opposing posterior teeth, the presence or absence of a posterior crossbite was listed as N/A. Additionally, if the participants were missing all their anterior teeth, the presence of anterior crossbite and anterior open bite was listed as N/A. The OHIP-14 scores for each of the seven domains and the overall OHIP-14 score were quantified and categorized by intraoral findings and are listed in Table 3.

	Posterior Crossbite	*Anterior Crossbite*	*Missing 8+ Teeth*	*Anterior Open bite*	*Prosthodontics and no orthodontics*
No	23	41	46	49	49
Yes	31	14	15	6	6
N/A	7	6	0	6	6 (Neither)

Table 2: Oral findings of the 61 individuals with CCD over the age of 15 that underwent an oral examination and completed the OHIP-14 survey.

The mean total OHIP-14 scores and mean individual domain scores for individuals with posterior crossbites, anterior open bites, anterior crossbites, missing 8+ teeth, and a history of prosthodontics without orthodontics were all higher (implying worse OHQOL) than their counterparts without the malocclusion or those who had orthodontics (Table 3 and Figure 13).

	Total OHIP	*Functional limitation*	*Physical pain*	*Psychological discomfort*	*Physical disability*	*Psychological disability*	*Social disability*	*Handicap*
Posterior Crossbite								
No	16.13	1.83	2.43	4.13	1.65	3.00	1.43	1.70
Yes	20.84	2.06	3.97	4.90	2.65	3.45	2.00	1.81
Anterior Open Bite								
No	18.24	1.76	3.35	4.57	2.04	3.20	1.63	1.69
Yes	25.33	3.17	3.83	5.67	2.33	4.33	3.00	3.17
Missing 8+ teeth								
No	17.1	1.6	3.2	4.5	1.6	3.1	1.5	1.6
Yes	30.9	3.7	5	5.7	4.9	4.9	3.6	3.2
Anterior Crossbite								
No	17.98	1.59	3.27	4.56	1.95	3.12	1.73	1.78
Yes	22.08	2.85	3.62	5.08	2.46	4.00	1.85	2.23
Prosthodontics without Orthodontics								
No	18.82	2.02	3.29	4.69	1.96	3.33	1.76	1.80
Yes	29.67	2.83	5.33	6.00	4.67	4.67	3.50	2.67

Table 3: OHIP-14 results of the 61 individuals with CCD over the age of 15 that underwent an oral examination and completed the OHIP-14 survey. Data is separated by oral finding and both total OHIP-14 and individual OHIP-14 domain scores.

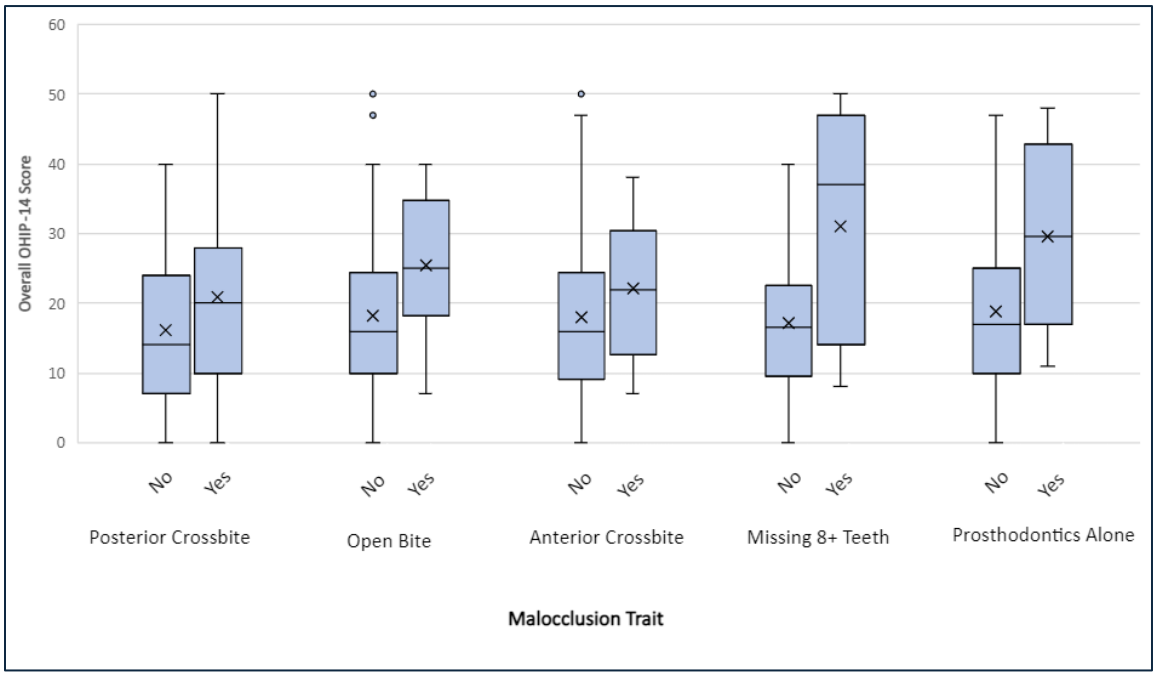


Figure 13: Box and Whisker which illustrates the distribution of total OHIP-14 scores. Total OHIP-14 scores were higher when there was a presence of posterior crossbite, open bite, or anterior crossbite. Those missing 8+ teeth had higher median total OHIP-14 scores than those with less than 8 missing teeth. Those who underwent prosthodontic treatment without orthodontics had higher median total OHIP-14 scores than those who underwent orthodontic treatment.

3.3 Statistical Analysis

In the comparison of samples for posterior crossbite, anterior open bite, anterior crossbite, and prosthodontics alone versus prosthodontics with orthodontics or orthodontics alone, Levene's test indicated non-significant results, allowing for the assumption of equal variances. However, since Levene's test indicated significant results for the comparison of samples of missing teeth, this oral finding was excluded from statistical analysis. Independent sample t-tests revealed that individuals with CCD who had posterior crossbites experienced significantly worse oral health-related quality of life (OHQOL) in the domain of physical pain ($p = 0.015$, 95% CI: [0.316, 2.750], Cohen's $d = 0.696$). Additionally, the domain of physical disability had a p-value which approached

significance (p = 0.091, 95% CI: [-0.164, 2.15], Cohen's d = 0.474). This suggests that these individuals endure higher levels of pain and physical impairment (Table 4).

For those with anterior open bites, a significantly higher OHIP-14 score was observed in the handicap domain handicap (p = 0.046, 95% CI: [0.027, 2.919], Cohen's d = 0.884). Additionally, the domains of functional limitation (p = 0.069, 95% CI: [-0.114, 2.937], Cohen's d = 0.803), and social disability (p = 0.079, 95% CI: [-0.165, 2.900], Cohen's d = 0.774) had results which approached significance (Table 5).

Furthermore, t-tests for individuals with anterior crossbites found statistically significant differences only in the functional limitation domain (p = 0.027, 95% CI: [0.149, 2.373]), with a medium to large effect size (Cohen's d = 0.724), indicating that these individuals faced more pronounced functional challenges (Table 6).

Lastly, independent t-tests demonstrated that individuals who received only prosthodontics reported significantly higher overall OHIP-14 scores (p = 0.037, 95% CI: [0.685, 21.015], Cohen's d = 0.926), indicating a worse OHQOL compared to those who received a combination of prosthodontics and orthodontics, or orthodontics alone. These patients also reported greater physical pain (p = 0.038, 95% CI: [0.114, 3.981], Cohen's d = 0.919), physical disability (p = 0.005, 95% CI: [0.872, 4.543], Cohen's d = 1.280), and social disability (p = 0.039, 95% CI: [0.091, 3.398], Cohen's d = 0.915), with large effect sizes in all cases (Table 7).

Domain	*Two-sided p-value*	*Difference in Means*	*95% Confidence Interval*	*Cohen's d*
OHIP-14 Score	0.161	4.708	[-1.929, 11.345]	0.392
Functional Limitation	0.653	0.238	[-0.820, 1.297]	0.124
Physical Pain	0.015*	1.533	[0.316, 2.750]	0.696
Psychological Discomfort	0.256	0.773	[-0.565, 2.111]	0.319
Physical Disability	0.091	0.993	[-0.164, 2.150]	0.474
Psychological Disability	0.444	0.452	[-0.724, 1.627]	0.212
Social Disability	0.261	0.565	[-0.432, 1.563]	0.313
Handicap	0.811	0.111	[-0.814, 1.035]	0.066

Table 4: P-values (*p < 0.05), differences in means, 95% CI, and effect size from t-test comparing OHIP-14 scores of those with CCD with posterior crossbite and those without.

Domain	*Two-sided p value*	*Mean Difference*	*95% Confidence Interval*	*Cohen's d*
OHIP-14 Score	0.180	7.088	[-3.366, 17.543]	0.558
Functional Limitation	0.069	1.412	[-0.114, 2.937]	0.803
Physical Pain	0.627	0.627	[-1.507, 2.480]	0.212
Psychological Discomfort	0.298	1.095	[-0.994, 3.184]	0.455
Physical Disability	0.755	0.293	[-1.581, 2.166]	0.135
Psychological Disability	0.236	1.129	[-0.760, 3.018]	0.519
Social Disability	0.079	1.367	[-0.165, 2.900]	0.774
Handicap	0.046*	1.473	[0.027, 2.919]	0.884

Table 5: P-values (*p < 0.05), differences in means, 95% CI, and effect size from t-test comparing OHIP-14 scores of those with CCD with anterior open bite and those without.

Domain	*Two-sided p value*	*Mean Difference*	*95% Confidence Interval*	*Cohen's d*
OHIP-14 Score	0.297	4.101	[-3.716, 11.919]	0.335
Functional Limitation	0.027*	1.261	[0.149, 2.373]	0.724
Physical Pain	0.636	0.347	[-1.117, 1.811]	0.151
Psychological Discomfort	0.510	0.516	[-1.046, 2.078]	0.211
Physical Disability	0.464	0.510	[-0.876, 1.897]	0.235
Psychological Disability	0.214	0.878	[-0.524, 2.280]	0.400
OHIP Social Disability	0.845	0.114	[-1.053, 1.282]	0.063
Handicap	0.414	0.450	[-0.646, 1.547]	0.262

Table 6: P-values (*p < 0.05), differences in means, 95% CI, and effect size from t-test comparing OHIP-14 scores of those with CCD with anterior crossbite and those without.

Domain	*Two-sided p value*	*Mean Difference*	*95% Confidence Interval*	*Cohen's d*
OHIP-14 Score	0.037*	10.85	[0.685, 21.015]	0.926
Functional Limitation	0.320	0.813	[-0.811, 2.437]	0.434
Physical Pain	0.038	2.048	[0.114, 3.981]	0.919
Psychological Discomfort	0.207	1.306	[-0.745, 3.358]	0.552
Physical Disability	0.005*	2.707	[0.872, 4.543]	1.280
Psychological Disability	0.134	1.34	[-0.428, 3.108]	0.658
Social Disability	0.039*	1.745	[0.091, 3.398]	0.915
Handicap	0.200	0.871	[-0.474, 2.215]	0.562

Table 7: P-values (*p < 0.05), differences in means, 95% CI, and effect size from t-test comparing OHIP-14 scores of those with CCD who underwent prosthodontics and those who underwent orthodontics alone or orthodontics in combination with prosthodontics

3.4 Other Findings- Orthodontics vs Prosthodontics

There were 23 members of CCD Smiles aged 18 and up who had orthodontic treatment and responded to the questions of "How many years were you in orthodontic treatment?" and "Do you wish you had gotten prosthetic (fake) teeth rather than orthodontics to bring your own teeth into the mouth?" (APPENDIX C). Of these 23 individuals, 19 had completed their orthodontic treatment. These patients averaged 9.2 +/- 5.5 years of orthodontic treatment with 2 patients having orthodontic treatment last for over 20 years (Figure 14). Nine of these twenty-three individuals stated that they would rather have received prosthodontics alone instead of orthodontics. Reasons given were "orthodontics took too long" (5 individuals), "the exposures were painful" (1 individual), "the orthodontist did not know enough about the condition" (1 individual), "not being satisfied with the outcome" (1 individual), and that "since prosthodontics was needed anyways, it may have been best to receive prosthodontics alone" (1 individual).

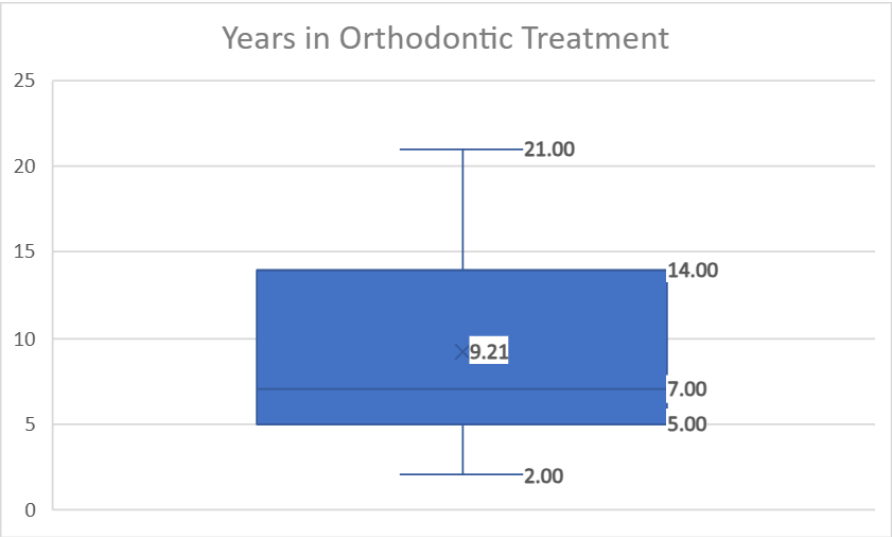


Figure 14: Distribution of number of years in orthodontic treatment of those with CCD who completed their orthodontic treatment.

3.5 Other Findings- Unilateral vs Bilateral Crossbite

When analyzing the differences between the OHIP-14 results between those with unilateral and bilateral crossbites, those with bilateral crossbites had a worse overall OHIP-14 score and worse scores in each of the 7 domains aside from the domain of social disability. Those with unilateral crossbites had similar OHIP-14 scores as those without crossbites in the domains of functional limitation, psychological disability and handicap (Table 8).

OHIP-14 scores for Crossbite

	Total no.	*Total OHIP*	*Functional limitation*	*Physical pain*	*Psychological discomfort*	*Physical disability*	*Psychological disability*	*Social disability*	*Handicap*
Neither	23	16.1	1.8	2.4	4.1	1.7	3	1.4	1.7
Unilateral	17	19.6	1.8	3.9	4.6	2.5	3.1	2.1	1.7
Bilateral	14	22.3	2.4	4.1	5.2	2.9	3.9	1.9	1.9

Table 8: Comparison of OHIP-14 scores of unilateral vs. bilateral vs. no crossbite.

3.6 Other Findings- Missing Teeth, Anterior vs. Posterior

The OHIP-14 results among those individuals missing teeth were higher than those not missing teeth in all categories aside from “Handicap” in which those missing posterior teeth only had a lower OHIP-14 score (1.2 to 1.4). Those missing anterior teeth only or both anterior and posterior teeth, had worse OHIP scores across all domains (Table 9).

OHIP-14 scores for Missing Teeth

	Total	*Total OHIP*	*Functional limitation*	*Physical pain*	*Psychological discomfort*	*Physical disability*	*Psychological disability*	*Social disability*	*Handicap*
Neither	10	12.9	1.3	2.6	3.1	1.1	2.3	1.2	1.4
Anterior only	2	34	4	6	6	4	6	4	4
Posterior only	18	16.3	1.1	3.2	4.9	1.6	3	1.4	1.2
Both	30	24.1	2.8	4	5.1	3.2	4.0	2.5	2.5

Table 9: Comparison of OHIP-14 scores based on category of missing teeth

4. Discussion

This study compared oral exam findings to OHQOL in individuals with CCD. There have been studies which described the intraoral and extraoral features of CCD[2,11,31,33], however, there are no known studies which investigate if oral findings in individuals with CCD affect their OHQOL. Additionally, there is no known study which compares the OHQOL of individuals with CCD who received orthodontic treatment to those who received prosthodontics alone to treat their condition. The discussion will begin with the results from the oral exam findings then proceed to discuss the OHIP-14 results and how certain malocclusions and oral findings affect OHQOL.

4.1 Oral Exam Findings

The intraoral and extraoral examinations of 75 individuals over 15 years of age revealed that a majority had brachyfacial facial types, high arch palates, and almost half had concave profiles. These characteristics have been reported in the literature. [2,3,11,33] In our study, 70.6% of affected individuals over 15 years of age were classified as being brachyfacial. This is well above the 46.5% reported in systematic reviews from South America.[26] The disparity in these findings may be due to the homogenous ethnicities in the subjects examined in the South American study.

The concave profile seen in individuals with CCD is usually due to maxillary hypoplasia and relative mandibular prognathism.[2,3,11,33] Interestingly, in a study regarding the craniofacial morphology of 28 Japanese individuals with cleidocranial dysplasia, they determined that these subjects' maxillae were not hypoplastic and were either averaged or

above averaged sized, and their mandibles were increased in size.[33] In this study, 44% of those examined were classified as having concave profiles. However, we did not have access to cephalometric measurements to classify these individuals as having hypoplastic maxillae, prognathic mandibles, or a combination of both.

Our oral examinations also revealed a high percentage of the examinees had anterior crossbites (23%), posterior crossbites (49%), Angle's class III molar relationship (47%), and open bites (11%). All these findings may be due to maxillary hypoplasia, a common finding in those with CCD,[11] which can manifest itself in all three dimensions.[34] Maxillary hypoplasia in an anterior-posterior dimension can cause anterior crossbites and result in Angle's class III molars, maxillary hypoplasia in a transverse dimension can result in unilateral and bilateral posterior crossbites, and maxillary hypoplasia in a vertical dimension can cause anterior open bites.[35] Maxillary hypoplasia may also result in a deep bite due to autorotation of the mandible against a deficient maxilla.[34]

The percentages of each type of malocclusion seen in our sample in patients with CCD are higher than the average sample of the public. A 2018 systematic review which looked at the global distribution of malocclusion traits found that in the permanent dentition, 4.93% of patients had open bites, 9.39% of individuals had posterior crossbites, and 5.93% had Angles class III malocclusion.[36] The NHANES III survey, a survey which examined the prevalence of malocclusion and orthodontic treatment need in the United States, found that 3.3% of individuals older than 18 had anterior open bites, 9.5% had posterior crossbites and 5.8% had zero or negative overjet.[37] Although the percentages of each of these malocclusions (11% for anterior open bite, 23% for anterior crossbite, and 49% for post crossbite) found in our sample are higher than the average, it is probable that

these percentages would be even higher had none of the individuals received orthodontic intervention.

In the 2018 study by Impellizzeri et al., 100% of individuals with CCD had maxillary constriction which needed some form of dental or orthopedic intervention.[23] In our study, 80% of patients had high arched palates, and 49% had a posterior crossbite. There were only 4 patients out of the 75 examined that did not have either a posterior crossbite or high arched palate. Since 3 of those 4 patients had received orthodontic treatment, it is possible that almost all participants had a form of transverse discrepancy. This data is consistent with Impellizzeri et al's observation. However, in Impellizzeri's research, 98% of subjects with CCD had a class III dental relationship. This contrasts with our study which found that only 62% of those who had molar relationships had at least one Angle's class III molar. It is likely that our examinations revealed a reduced percentage of class III molars since many of the participants in our study had received orthodontic treatment in the past.

In our sample, 33.8% had 8 or more missing teeth. These individuals likely had a reduced OHQOL due to this. According to Gerritsen et al, when an individual is missing 8 or more teeth, there are negative implications to the individual's OHQOL. Specifically, missing this number of teeth leads to suboptimal masticatory function and even if these teeth are replaced, it is difficult to adapt to their replacements.[32]

4.2 OHIP-14 Surveys

Validated surveys have been used to determine an individual's oral health related quality of life. The OHIP-49 is one of these validated surveys.[38] This survey aims to

determine the oral health related quality of life across seven domains: Functional Limitation, Physical Pain, Psychological Discomfort, Physical Disability, Psychological Disability, Social Disability, and Handicap. The OHIP-14 is a shortened version of the OHIP-49.[39] Fourteen questions were selected from the OHIP-49 to create the OHIP-14 using regression analysis and factor analysis to determine which questions of the OHIP-49 were the most important in predicting the overall impact of oral health on quality of life and to ensure that the selected questions covered all relevant aspects of oral health. Internal Reliability Analysis yielded a Cronbach's Coefficient Alpha of 0.88, showing that the OHIP-14 questions are very consistent in measuring the impact of oral health on quality of life. Shortening the survey from 49 to 14 questions reduces the likelihood of survey fatigue and increases the chances of survey completion.[40]

A systematic review with meta-analysis used studies which used the OHIP-14 survey to determine the impact of malocclusion on people's quality of life. The review concluded OHIP-14 scores were higher in those with malocclusion than those without malocclusion, and that orthodontic treatment significantly improves oral health related quality of life.[41]

4.3 OHIP-14 results

Of the 75 individuals who completed the oral examination, 61 also completed the OHIP-14 survey. These survey results showed that individuals with CCD who had prosthodontic treatment without orthodontic treatment, the presence of anterior crossbite, posterior crossbite, anterior open bite, or having eight or more missing teeth had higher

OHIP-14 scores in comparison to those with CCD without these findings. Although the OHIP-14 scores were lower in individuals with CCD without these findings, the total OHIP-14 scores were still high and ranged from 16 to almost 19. These high scores indicate that even those with CCD who had orthodontic treatment or did not have an anterior crossbite, posterior crossbite, anterior open bite, or eight or more missing teeth, have poor OHQOL. According to Roumani et al, OHIP-14 scores of over 11 indicate poor oral health which has a significant impact on quality of life.[42]

For the comparisons involving posterior crossbite, anterior open bite, anterior crossbite, and prosthodontic versus orthodontic treatment modalities, Levene's test confirmed the assumption of equal variances, supporting the robustness of the t-test results. However, the significant results for Levene's test in the analysis of missing teeth warranted the exclusion of this variable from statistical analysis due to potential violation of homogeneity assumptions.

In individuals with posterior crossbites, a significant impairment in the domain of physical pain highlights the tangible burden these patients experience. The moderate to large effect sizes (Cohen's d = 0.696) suggest that this finding is not only statistically significant but also clinically meaningful. This impairment in the domain of physical pain is likely due to occlusal imbalances and strain on the masticatory system.[43]

For individuals with anterior open bites, a significant impact was observed in the domain of handicap and approached significance in the domains of functional limitation and social disability. The large effect sizes (Cohen's d ranging from 0.774 to 0.884) indicate profound effects on both functional and psychosocial aspects of quality of life. These

findings underscore the multifaceted challenges faced by individuals with anterior open bites, which may include difficulties with speech, mastication, and social interactions.[44] Such results reinforce the importance of comprehensive treatment approaches that address both the physical and social implications of this malocclusion.

In the case of anterior crossbites, significant differences were limited to the functional limitation domain, with a medium to large effect size (Cohen's d = 0.724). This suggests that while anterior crossbites may not affect all dimensions of quality of life, they specifically impair functional capacity, likely due to challenges with mastication.[45]

The comparison of treatment modalities revealed that individuals with CCD receiving only prosthodontic care reported significantly worse OHQOL across multiple domains, including physical pain, physical disability, and social disability. The large effect sizes observed (Cohen's d ranging from 0.915 to 1.280) suggest a substantial benefit of incorporating orthodontic treatment, either in isolation or in combination with prosthodontics.

Prosthodontic treatment alone was associated with poorer social outcomes and more physical pain and disability. While prosthodontics restores missing teeth, it may fail to replicate the aesthetics and alignment of natural teeth, leading to self-consciousness and social discomfort during speaking or smiling. Additionally, prosthetic teeth often do not correct underlying malocclusions, which can result in uneven masticatory forces, TMJ strain, and chronic pain. Ill-fitting devices, such as dentures, can cause gum irritation or oral sores, increasing physical discomfort and negatively impacting social engagement.

Fear of prosthetic slippage during eating or speaking further contributes to social anxiety, discouraging social interactions.

In contrast, orthodontic treatment aligns natural teeth, enhancing aesthetics, restoring proper occlusion, and reducing areas of excessive strain, which alleviates pain and encourages confidence in social settings. These findings emphasize the importance of orthodontics in improving long-term oral health, physical comfort, and social quality of life, beyond what prosthodontics alone can achieve.

Since individuals with CCD who have an anterior crossbite, posterior crossbite, anterior open bite, or had prosthodontic treatment without orthodontic treatment had statistically significantly higher OHIP-14 scores in comparison to those with CCD without these findings in specific areas, the null hypotheses are rejected.

Overall, these findings emphasize the significant impact of malocclusions and treatment modalities on OHQOL in individuals with CCD. The results underscore the need for treatment to optimize outcomes and address both the physical and psychosocial dimensions of quality of life.

4.4 Additional Findings

This study offers important insights into the experiences of adults with CCD who have undergone orthodontic treatment. Among 23 individuals who completed treatment,

the average duration was remarkably long, at 9.2 ± 5.5 years, with two patients undergoing treatment for over 20 years. Despite completing their orthodontic care, nearly 40% indicated they would have preferred prosthodontic replacement of their impacted teeth, citing lengthy treatment times, pain from surgical exposures, dissatisfaction with outcomes, and insufficient orthodontic clinician expertise in managing CCD. This feedback underscores the need for more efficient treatment protocols and enhanced provider education in addressing CCD-related malocclusions. However, based on OHIP-14 results, it appears that while a significant proportion of patients may have expressed a preference for prosthodontics, their overall oral health-related quality of life was ultimately better with orthodontic treatment. These findings emphasize the long-term benefits of orthodontics despite the challenges patients faced during treatment. Additionally, these findings underscore the need to educate individuals with CCD about the long-term benefits of orthodontic treatment to help them reshape their expectations and perceptions and give them the opportunity to make informed decisions

Analysis of OHIP-14 scores revealed that bilateral crossbites significantly impaired OHQOL, with worse scores across most domains compared to those with unilateral crossbites and those without crossbites. Notably, individuals with unilateral crossbites showed comparable scores to those without crossbites in the domains of functional limitation, psychological disability, and handicap, suggesting that bilateral crossbites present greater challenges in both physical and psychological dimensions than unilateral crossbites.

Missing teeth had a pronounced impact on OHQOL, particularly in those missing anterior teeth or both anterior and posterior teeth, who exhibited worse scores across all

domains in comparison to those missing posterior teeth only and to those not missing any teeth. These findings underscore the importance of preserving natural teeth, particularly in the anterior region, to minimize functional and psychosocial impacts.

5. Limitations

This study is limited by the relatively small number of participants, which may reduce the robustness of the statistical analyses. However, considering that CCD is a rare condition, occurring in approximately one in a million individuals, this sample size of 75 participants for oral exams and 61 for statistical analysis represents a significant opportunity for research. Additionally, the oral examination results may be less conclusive, as a high percentage of participants had undergone orthodontic treatment, potentially influencing the findings. Furthermore, when comparing orthodontics to prosthodontics, this study did not differentiate whether patients received implants or dentures to replace their missing teeth. It is possible that if we compared the OHQOL of individuals who received implants without having orthodontic treatment, the OHQOL would be similar.

6. Future Research

Future research should focus on using regression analysis to correlate oral health findings with OHIP-14 scores to identify more detailed relationships between clinical conditions and quality of life. This could enhance the understanding of CCD's impact on

oral health and guide more tailored treatment approaches. Additionally, it would be beneficial in the future to compare the OHQOL of individuals who received implants to those who received removable prosthodontics. Lastly, we hope to obtain cephalometric data, to help establish cephalometric norms for CCD.

6. Conclusion

In conclusion, individuals with CCD who have anterior crossbites, posterior crossbites, or anterior open bites have statistically significantly worse OHQOL in specific domains comparison to those with CCD without these findings. Additionally, although 40% of individuals with CCD who had orthodontic treatment stated that they would have preferred prosthodontic replacement of impacted teeth, OHIP-14 results indicate that those who had prosthodontics alone had statistically significant worse OHQOL than those who had orthodontics in the domains of physical pain, physical discomfort and social disability. Overall, these findings emphasize the significant impact of malocclusions and treatment modalities on OHQOL in individuals with CCD. These results underscore the need for treatment to optimize outcomes and address both the physical and psychosocial dimensions of quality of life.

APPENDIX A

Oral Exam Form

Date collected: __ __ / __ __ / __ __ __ __
Name of examiner: _____
Participant ID: _ __ __ __ - __ __ __
DOB: __ __ / __ __ / __ __ __ __
SEX: female male

1. **Facial profile**: convex / concave / straight
2. **Facial length**: dolicofacial / brachyfacial / mesofacial
3. **Asymmetry**: yes / no
4. **Palate**: cleft palate / normal / high arched
5. **Teeth Present:**

1 2 3 4 5 6 7 8 / 9 10 11 12 13 14 15 16 A B C D E / F G H I J
32 31 30 29 28 27 26 25 / 24 23 22 21 20 19 18 17 T S R Q P / O N M L K

6. **Molar classification:**

Side	class I	½ cusp class II	full-cusp class II	½ cusp class III	full-cusp class III
Right					
Left					

7. **Anterior crossbite**: yes / no
8. **Posterior crossbite**: unilateral / bilateral / none
9. **Overbite**: ____mm
10. **Overjet**: ____mm
11. **Arch length:**

	Crowding			Spacing			Normal
Arch	Mild (1-3mm)	Moderate (4-6mm)	Severe (7mm+)	Mild (1-3mm)	Moderate (4-6mm)	Severe (7mm+)	
Maxillary							
Mandibular							

12. **OH/caries**: good OH / fair OH / poor OH / overt caries present / white spots present
13. **Perio**: normal / recession on _____ teeth
14. **Braces on**: yes / no
    a. Teeth exposed: _____
15. **Prosthetic teeth**: yes / no / which teeth and prosthesis: _____ ____

_____

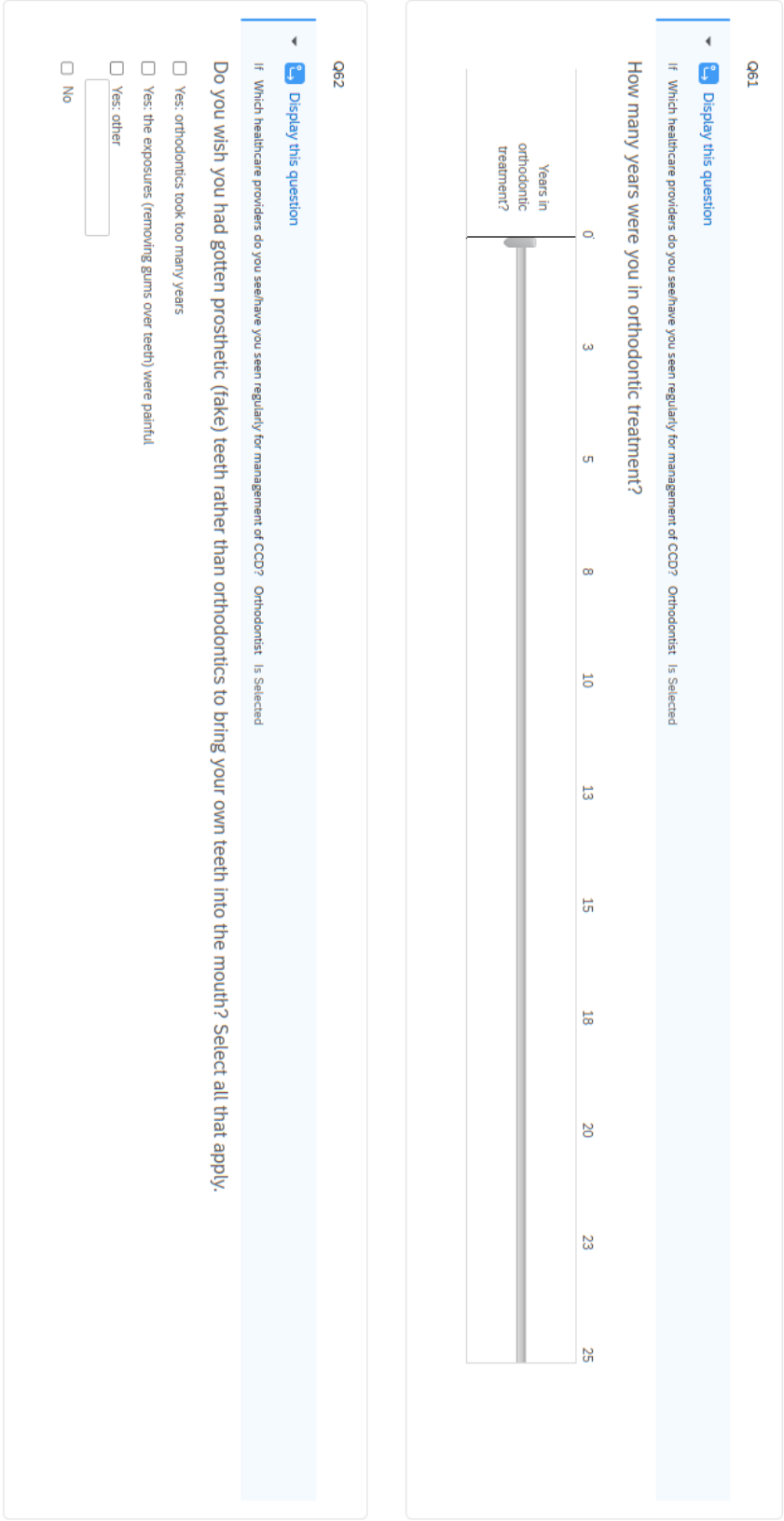
APPENDIX B

OHIP-14

During the last 12 months, how often has the following occurred?

	Never	Hardly Ever	Occasionally	Fairly Often	Very Often
Have you had trouble pronouncing any words because of problems with your teeth, mouth or dentures?	○	○	○	○	○
Have you felt that your sense of taste has worsened because of problems with your teeth, mouth or dentures?	○	○	○	○	○
Have you had painful aching in your mouth?	○	○	○	○	○
Have you found it uncomfortable to eat any foods because of problems with your teeth, mouth or dentures?	○	○	○	○	○
Have you been self-conscious because of your teeth, mouth or dentures?	○	○	○	○	○
Have you felt tense because of problems with your teeth, mouth or dentures?	○	○	○	○	○
Has been your diet been unsatisfactory because of problems with your teeth, mouth or dentures?	○	○	○	○	○
Have you had to interrupt meals because of problems with your teeth, mouth or dentures?	○	○	○	○	○
Have you found it difficult to relax because of problems with your teeth, mouth, or dentures?	○	○	○	○	○
Have you been a bit embarrassed because of problems with your teeth, mouth, or dentures?	○	○	○	○	○
Have you been a bit irritable with other people because of problems with your teeth, mouth, or dentures?	○	○	○	○	○
Have you had difficulty doing your usual jobs because of problems with your teeth, mouth, or dentures?	○	○	○	○	○
Have you felt that life in general was less satisfying because of problems with your teeth, mouth, or dentures?	○	○	○	○	○
Have you been totally unable to function because of problems with your teeth, mouth, or dentures?	○	○	○	○	○

APPENDIX C


Q61
Display this question
If Which healthcare providers do you see/have you seen regularly for management of CCD? Orthodontist Is Selected
How many years were you in orthodontic treatment?
Years in orthodontic treatment?
0
3
5
8
10
13
15
18
20
23
25
Q62
Display this question
If Which healthcare providers do you see/have you seen regularly for management of CCD? Orthodontist Is Selected
Do you wish you had gotten prosthetic (fake) teeth rather than orthodontics to bring your own teeth into the mouth? Select all that apply.
Yes: orthodontics took too many years
Yes: the exposures (removing gums over teeth) were painful
Yes: other
No

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