Oral Health-Related Quality of Life in Children with Oligodontia

by

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A thesis submitted in conformity with the requirements for the degree of Master of Science Pediatric Dentistry
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Abstract

**Background:** Oligodontia (congenital absence of six or more teeth) affects functional, emotional and social aspects of an individual’s life. Few published studies have evaluated oral health-related quality of life (OHRQoL) in children with oligodontia.

**Methods:** Thirty-five 8-to-18-year-old patients with oligodontia recruited from The Hospital for Sick Children and Holland Bloorview Kids Rehabilitation Hospital, and their parents, completed the short format of Child Perception Questionnaire (CPQ11-14) and Parent Child Perception Questionnaire (P-CPQ), respectively.

**Results:** Children reported significantly higher overall CPQ score than their parents. Correlations between children’s and parents’ overall CPQ score, oral symptoms and functional limitations, and social well-being were not statistically significant. However, as children’s emotional well-being score increased, parents’ score also increased. There was no association between child CPQ score and age, gender, number and location of permanent tooth agenesis in this sample.

**Conclusion:** Children with oligodontia had poorer OHRQoL compared to what was perceived by their parents.
Dedicated to my daughter Lianna.
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**List of Abbreviations**

OHRQoL: Oral Health-Related Quality of Life

P-CPQ: Parental-Caregiver Perception Questionnaire

ED: Ectodermal Dysplasia

OFD I: Oral-Facial-Digital Syndrome Type I

VWS: Van Der Woude Syndrome

GOHAI: Geriatric/General Oral Health Assessment Index

OHIP: Oral Health Impact Profile

COHIP: Child Oral Health Impact Profile

OIDP: Oral Impacts on Daily Performances

CPQ: Child Perception Questionnaire

CPQ8-10: Child Perception Questionnaire for 8- to 10-year-old children

CPQ11-14: Child Perception Questionnaire for 11- to 14-year-old children

C-OIDP: Child Oral Impacts on Daily Performances

ECOHIS: Early Child Oral Health Impact Scale

SOHO-5: Scale of Oral Health Outcomes for 5-year-old children

MOHRQoL: Michigan Oral Health-Related Quality of Life Scale

POQL: Pediatric Oral Health- Related Quality of Life Measure

Coopersmith SEI-SF: Coopersmith Self-Esteem Inventory-School Form

VAS: Visual Analogue Scale

CLP/CD: Cleft Lip and Palate/Craniofacial Dental Program

MOHLTC: Ontario Ministry of Health and Long-Term Care
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Chapter 1

Introduction

Teeth play an important role in our day-to-day lives. Their appearance, including shape, color, proportion and positioning, contributes to the esthetic determinants that can affect an individual’s personality. The dentition also serves important functions in mastication, nutrition and speech. Dental agenesis, depending on severity, can affect both esthetic determinants and important sensorimotor functions of the oral cavity. While agenesis of one or a few teeth (hypodontia) is relatively common, an increased severity of dental agenesis is rare.

Oligodontia is defined as the congenital absence of six or more permanent teeth, excluding third molars and can have adverse effects on functional, emotional and social aspects of a patient’s life (1). The prevalence of hypodontia varies among different study populations (2.6-11.3%) (2,3) whereas oligodontia is much more rare, with prevalence reported between 0.03 and 0.07% (1,4). Optimal dentofacial management of affected patients requires a multidisciplinary treatment approach including pediatric dentistry, orthodontics, prosthodontics and oral and maxillofacial surgery (5).

A review of the literature demonstrated that only a few studies have evaluated oral health-related quality of life (OHRQoL) in children with hypodontia and oligodontia. Studies using validated questionnaires have shown that oligodontia can have negative functional and psychological impact on patients at different ages of their childhood (6–8). Parental report of their children’s OHRQoL have not necessarily been correlated or correlated strongly with the impacts that have been demonstrated in affected children (9). Consequently, there is a lack of a clear understanding of whether oligodontia affects OHRQoL in affected children and their parents.
Comparing the child’s input with parent’s input report is important in understanding these impacts and developing a comprehensive treatment plan to address them in an optimal manner. It is also important to determine if other factors such as age, sex, and socioeconomic status impact the oral health-related quality of life in these patients.

This thesis describes an investigation that assessed the OHRQoL in children with oligodontia, determined the parents’ views regarding OHRQoL of their children with oligodontia, analyzed the correlation between children’s and parents’ reported OHRQoL of affected children, and evaluated the potential effects of the number and location of missing teeth, children’s age, sex and family income on OHRQoL of affected children.
Chapter 2

Review of the literature

2.1 Definition

Tooth agenesis is defined as the developmental absence of teeth. It is one of the most common human congenital dental anomalies. Tooth agenesis has three different expressions: hypodontia, oligodontia, anodontia. Hypodontia is congenital absence of five or fewer teeth, excluding the third molars. Oligodontia is congenital absence of six or more teeth, excluding the third molars (3–5). Anodontia is congenital absence of all teeth (4,5).

2.2 Prevalence and Location

The prevalence of hypodontia geographically has been reported to vary among populations studied. It has been reported to be 2.7% among Mexicans (10), 4.3%-7.54% in the Turkish population (4,11), 4.19% in the Indian population (3), 4.5% among the Norwegians (12), 5.21% among Iranians (2), 6.3 % in the Brazilian population (13), 8.5% in the Japanese population (14) and 11.6% among Koreans (15). The reported prevalence of hypodontia in primary dentition is lower than permanent dentition. A study by Magnússon et al. in Iceland showed the prevalence of 0.5% for hypodontia in primary dentition (16). Tooth agenesis is more common in black Americans than in white Americans (17). The prevalence of tooth agenesis is higher in Europe and Australia than in North America (18). The prevalence of oligodontia is lower than that of hypodontia and varies among different study populations: 0.084% among Norwegians (12), 0.09% in the Swedish population (19), 0.07, 0.3% and 1.13% in the Turkish population (1,4,20), 0.16% in the Danish population (21), 0.34% in the Iranian population (22), and1.04% in the Hungarian population (23).
Tooth agenesis is more common in females than in males (1,4,11,12,18,21,25–26). In a review article by Larmour et al. A female-to-male ratio of 3:2 was reported for hypodontia (26). According to the meta-analysis by Polder et al. (18), females had 1.37 times higher prevalence of tooth agenesis compared to males.

Tooth agenesis in the permanent dentition is more common in the maxilla compared to the mandible (1,4,11). In the permanent dentition, the prevalence of bilateral tooth agenesis is more common than unilateral tooth agenesis(1). The most commonly missing permanent teeth are third molars with a prevalence of 24% (10).

The most commonly missing permanent teeth, excluding the third molars are mandibular second premolars, maxillary lateral incisors, maxillary second premolars and mandibular central incisors in different orders depending on the study (2,4,11,12,24). In a study performed by Celikoglu et al., the most prevalent tooth agenesis in patients undergoing orthodontic treatment were maxillary lateral incisors, mandibular second premolars and mandibular central incisors in descending order (4). The most frequent missing permanent teeth in a study by Sisman et al. were maxillary lateral incisors, mandibular second premolars and maxillary second premolars (11). The permanent teeth more often missing in a study by Nordgarden et al. were the lower second premolars, maxillary lateral incisors and maxillary second premolars (12).

The most commonly missing primary teeth are maxillary lateral incisors (27) and mandibular lateral incisors (28). Unilateral tooth agenesis was more common in the primary dentition than bilateral tooth agenesis (28).
2.3 Etiology

Non-Syndromic Hypodontia:

Genetic factors: Brooks et al. proposed a multifactorial model including both environmental and polygenic components to explain anomalies of tooth size (microdontia, macrodontia) and number (hypodontia, supernumeraries) (29). Relatives of affected individuals were significantly more affected compared to the general population. Individuals with more severe hypodontia were more likely to have affected relatives. Having more than one affected individual in a family increased the risk of having affected relatives (29).

Genes responsible for non-syndromic hypodontia are MSX1 (causing oligodontia) and PAX9 (causing agenesis of molars, maxillary lateral incisors and premolars). AXIN2 can cause a mixed pattern of agenesis. FGFR1 causes agenesis in the premolar area. TGFA plays a role in incisor agenesis (30).

Parkin et al. reported that tooth agenesis was significantly more common in siblings and parents of affected individuals, but that there was no significant relationship for the site and number of missing permanent teeth between parents or siblings and affected individuals. This study also confirmed the multifactorial etiology of hypodontia with genetic, epigenetic and environmental factors playing a role (31).

Environmental factors: There is a lack of clear understanding of the role environmental factors play in tooth agenesis. According to the literature there are some environmental factors that can cause tooth agenesis:

Allergy: Yamaguchi et al. (32) showed that there is a positive significant relationship between allergy and hypodontia in orthodontic patients in Japan.
Infections: Some infections such as rubella can cause hypodontia (33).

Medications: Thalidomide taken by mothers during their pregnancy can cause tooth abnormalities including hypodontia (31,34).

Chemotherapy and radiation therapy: Children undergoing treatment at a young age can have hypodontia specially affecting premolars and molars (35,36)

**Syndromic Hypodontia:**

Tooth agenesis can present as part of some syndromes. Only a few well-known syndromes are included in this review to describe their patterns of tooth agenesis.

**Ectodermal dysplasia (ED)**

ED occurs in 1/100,000 live births. Ectodermal dysplasia is a group of conditions characterized by abnormal development of two or more ectodermally-derived tissues including the skin, hair, nails, teeth, sweat glands, salivary glands and distinctive craniofacial features such as frontal bossing, pronounced chins and broad noses. The most common form of ectodermal dysplasia is hypohidrotic ectodermal dysplasia (HED) which is characterized by sparse scalp and body hair (hypotrichosis), missing teeth (oligodontia), cone-shaped teeth, and absence of sweat glands (hypohidrosis). The inheritance pattern of HED is mostly X-linked recessive. (37,38). All the affected individuals present with either hypodontia or oligodontia. The most common missing teeth in ED in a study by Präger (39) were maxillary lateral incisors and mandibular central incisors.
Oral-facial-digital syndrome Type I (OFD I)

The incidence of OFD I is 1/50,000-250,000 live births. The most common pattern of inheritance is X-linked dominant. The clinical features of the syndrome include hypertelorism, syndactyly, polydactyly, cleft lip/palate, tongue abnormalities including bifid tongue, hyperplastic frenum, and missing teeth (40). The most commonly missing teeth in this syndrome are mandibular central incisors (41).

Down Syndrome

Down syndrome is one of the most common chromosomal abnormalities in humans. The most common oral characteristics of patients with Down syndrome or trisomy 21 are midface deficiency, relative mandibular prognathism, relative large tongue, fissured tongue, hypodontia, microdontia, supernumeraries, enamel hypoplasia, taurodontism and periodontal disease (42,43).

The prevalence of hypodontia in a sample of Down syndrome patients was 92% including the third molars and 56% excluding the third molars (44). The results of a meta-analysis by Palaska and Antonarakis showed that the prevalence of tooth agenesis was 54.6% excluding the third molars in patients with Down syndrome (42). According to the study by Kumasaka in Japan (45), the prevalence of oligodontia in patients with Down syndrome was 63% and the prevalence of hypodontia was 53%. The most common missing permanent teeth in these patients were mandibular lateral incisors (23.3%), maxillary second premolars (18.2%), maxillary lateral incisors (16.5%) and mandibular second premolars (15.3%).
Non-syndromic Cleft lip/palate

The most common dental findings in patients with cleft lip/palate are hypodontia, supernumeraries, transposition, enamel hypoplasia, dilaceration, microdontia, taurodontism, ectopic eruption and rotation (43). More severe types of cleft have higher prevalence of hypodontia (46). Missing of lateral incisor in the cleft area has been reported in 49.8% of the cases with unilateral cleft lip/palate. The most common tooth agenesis outside the cleft area is maxillary second premolar (47). In a study by Laatikainen et al. (48) 37% of monozygotic twins and 32.7% of dizygotic twins with cleft lip/palate had hypodontia. The most common tooth agenesis in their study were maxillary second premolar, maxillary lateral incisor, and mandibular second premolar in descending order.

Pierre-Robin sequence

The most common features of Pierre-Robin sequence are micrognathia, cleft palate and glossoptosis leading to airway obstruction in neonatal period (49,50). The prevalence of permanent tooth agenesis was higher in patients with Pierre-Robin sequence compared to patients with non-syndromic cleft palate (47.8% and 29.8%, respectively) (50). In a study by Antonarakis and Suri in Canada, the prevalence of tooth agenesis in patients with Pierre-Robin sequence was 32.9%. The most common missing permanent teeth were mandibular second premolars, maxillary second premolars, and maxillary lateral incisors in descending order (49).

Van Der Woude syndrome (VWS)

The characteristic features of VWS are cleft lip, cleft palate, paramedian lip pits and hypodontia. The inheritance pattern is autosomal dominant (30,51). Hypodontia is reported in 10-81% of
patients with VWS (51). The most common missing teeth outside of the cleft area in patients with VWS with bilateral cleft lip/palate were maxillary right second premolar followed by mandibular left second premolar, and mandibular second premolars in unilateral cleft lip/palate (52).

Dental anomalies commonly seen in patients with hypodontia:

Microdontia and conical teeth: small tooth size has been associated with hypodontia (53). According to the study by Brooks et al. (29) there is an association between hypodontia and microdontia. As the severity of hypodontia increases, the possibility of microdontia also increases. All the patients with six or more missing teeth in their study had microdontia. In a study by Lai et al. 8.9% of patients with hypodontia have conical incisors (54).

Ankylosed primary teeth: it was reported in 65.7% of patients with hypodontia (54).

Transposition of canine and first premolar: there is a strong association between hypodontia and transposition of maxillary canine and first premolar. In a study by Camilleri et al., 20% of patients with missing maxillary lateral incisors and 24% of patients with missing mandibular second premolars had transposition of maxillary canines and first premolars (55).

Taurodontism: the results of a study by Kan et al. showed that the prevalence of taurodontism in the first permanent molar was higher in girls with hypodontia compared to a control group without hypodontia (56). In a study by Lai et al. taurodontism of the first permanent molar was reported in 34.3% of cases with hypodontia(54).

Enamel hypoplasia: enamel hypoplasia a common finding in patients with hypodontia. In a study by Lai et al. it was reported in 11.9% of patients with hypodontia (54).
2.4 Impacts of Oligodontia

Oligodontia may cause functional and psychological impacts on the affected individuals. These patients can have esthetics concerns (2,53,57), spaced dentition (57,58) and issues with function including eating and speech (53,59). Oligodontia can also have negative impacts on the oral health-related quality of life of the patients (6,9,60).

Patients with oligodontia often require a multidisciplinary treatment approach including pediatric dentistry, orthodontics, prosthodontics, oral and maxillofacial surgery and even a geneticist (26). Oligodontia can have adverse effects on functional, emotional and social aspects of a patient’s life.

2.5 Oral Health-Related Quality of Life (OHRQoL)

According to Locker and Allen, Oral Health-Related Quality of Life (OHRQoL) is defined as “The impact of oral disorders on aspects of everyday life that are important to patients and persons, with those impacts being of sufficient magnitude, whether in terms of severity, frequency or duration, to affect an individual’s perception of their life overall.”(61).

There are different measures to assess OHRQoL including Geriatric/General Oral Health Assessment Index (GOHAI), Oral Health Impact Profile (OHIP), Oral Impacts on Daily Performances (OIDP), Child Perception Questionnaire (CPQ) (61).

2.6 Children Self-Reported OHRQoL and CPQ

A review of the literature demonstrated that very few reported studies have evaluated oral health-related quality of life (OHRQoL) in children with oligodontia.
Historically, parents were the source of information for assessing quality of life of their children, however, validated questionnaires have now been developed to assess quality of life in children based on their self-report.

In a systematic review (62), Barbosa and Gaviao assessed the validity of children’s self-report on their OHRQoL. This systematic review included thirteen cross-sectional studies on either patients or general population which used the Child Perception Questionnaire (CPQ) for two different age groups (CPQ8-10 and CPQ11-14) and also Child-Oral Impacts on Daily Performances (Child-OIDP). The questionnaires used in all included studies showed good validity. They suggested that there are some factors that can affect the children’s perceived oral health including age, gender, race, education, culture, social deprivation, treatment opportunities and self-perceived treatment needs. There was no statistically significant difference in the overall CPQ score between genders. Emotional well-being domain score was higher in females compared to males.

There has been an increasing interest in assessing children’s OHRQoL in recent years. In another systematic review (63), Barbosa and Gaviao suggested that there is a relationship between OHRQoL and oral conditions. The questionnaires used by these authors were Child Perception Questionnaire (CPQ), Child-Oral Impacts on Daily Performances index (Child-OIDP) and Child Oral Health Impact Profile (COHIP). There was a relationship between oral health-related quality of life and different oral conditions including: dental caries, fluorosis, malocclusion, cleft lip and palate, hypodontia, and gingival issues. Other than clinical variables, there were different factors influencing the oral health-related quality of life including child development, social and environmental factors, and personal variables.
According to a study by Gilchrist (64), patient reported outcome measures (PROMs) have been recently used in dentistry. They give the patients the advantage of assessing their own quality of life and being involved in their care. They also reduce the observer bias. Different PROMs have been used to assess OHRQoL. Different valid and reliable ways to assess oral health-related quality of life in children have been introduced including Child Perceptions Questionnaire (CPQ), Child Oral Health Impact Profile (COHIP), the Child Oral Impacts on Daily Performances (C-OIDP), Early Child Oral Health Impact Scale (ECOHIS), the Scale of Oral Health Outcomes for 5-year-old children (SOHO-5), the Michigan Oral Health-Related Quality of Life scale (MOHRQoL) and the Pediatric Oral Health-Related Quality of Life Measure (POQL). The authors included the three most common instruments in their review: CPQ_{8-10}, CPQ_{11-14}, and short format of CPQ_{11-14}, COHIP and C-OIDP. Among these, CPQ_{11-14} was the most common measure used to assess OHRQoL in children. The authors confirmed the validity and reliability of all three instruments.

Jokovic et al. (65) developed the CPQ_{11-14}. The study group consisted of 11- to 14-year-old children with dental caries, orthodontic conditions and orofacial disorders (mostly cleft lip and/or palate). Their self-report questionnaire consisted of 37 questions in four different domains including oral symptoms (n=6), functional limitations (n=9), emotional well-being (n=9), and social well-being (n=13). The children were asked to answer the questions according to their experiences during the past three months. The response options were scored as: “Never” = 0; “Once/twice” = 1; “Sometimes” = 2; “Often” = 3; and “Every day/almost every day” = 4. The questionnaire also included two global rating questions: “Would you say that the health of your teeth, lips, jaws and mouth is...?” with answers ranging from “Excellent” = 0 to “Poor” = 5 and “How much does the condition of your teeth, lips, jaws or mouth affect your life overall?” with
answers ranging from “Not at all” = 0 to “Very much” = 5. The overall CPQ score was calculated by adding the response codes for all 37 items. They also calculated domain scores by adding response codes in each of the four different domains. The results of the study showed the questionnaire’s validity and reliability. It also showed that oral and orofacial conditions can have considerable effect on OHRQoL of children. The overall CPQ scores were as follows: orofacial group (27.0), orthodontic group (22.0), pedodontic group (19.0). The authors suggested longitudinal studies to assess longitudinal validity and responsiveness of the CPQ

Jokovic et al. (66) developed CPQ 8-10. Their study consisted of two groups of 8- to 10-year-old children with dental caries or with cleft lip and palate. The questionnaire included 25 questions in 4 different domains consisting of oral symptoms, functional limitations, emotional well-being, and social well-being. The children were asked to answer the questions according to their experiences during the past four weeks. The answers were coded as “never” = 0; “once/twice” =1; “sometimes” = 2; “often” = 3; and “everyday/almost every day” = 4. Overall scores and domain scores were calculated by adding response codes. The questionnaire also had two global rating questions: 1) “When you think about your teeth or mouth, would you say that they are...” with the answers ranging from “very good” =0 to “poor” =3, and 2) “How much do your teeth or mouth bother you in your everyday life?” with answers ranging from “not at all” = 0 to “a lot” = 3. The total score was 18.6 ±12.6. The frequency of children reporting one or two symptoms “everyday/almost every day” or “often” was 34% for oral symptoms, 24% for functional limitations, 16% emotional-wellbeing and 25% for social well-being. The overall CPQ score in the orofacial group was slightly higher (19.1) than in the pediatric dentistry group (18.6) but the difference did not reach the significance level. Children in the pediatric dentistry group reported
more oral symptoms but children in the orofacial group reported more issues in the functional limitations and social well-being domains. Children with caries had higher mean overall score (21.1) compared to caries-free children (14.7). There was a positive relationship between overall CPQ score and the number of decayed tooth surfaces. The authors showed the validity and reliability of the questionnaire.

Foster Page et al. (67) assessed the construct validity of CPQ$_{11-14}$ in a cross-sectional study in New Zealand. The sample of their study included 430 twelve-to-13-year old school children in two different groups: dental caries or malocclusion. In general, the mean overall CPQ score was 17.3 with a range of 0 to 103. The overall CPQ score and all domain scores were higher in females compared to males, but the difference was not significant except for the emotional well-being score which was significantly higher in females compared to males. Children with the highest DMFS quartiles had higher overall CPQ scores (21.8) and domain scores (5.7 for oral symptoms, 6.1 for functional limitations, 4.4 for emotional well-being and 5.6 for social well-being). Children with more severe types of malocclusion had higher overall CPQ scores (21.6), emotional well-being (4.9) and social well-being (5.7). The results of their study suggested that the construct validity of the Child Perception Questionnaire (CPQ$_{11-14}$) is acceptable in a sample of New Zealander children with more severe types of malocclusions and higher caries experience.

Wogelius et al. (68) developed Danish versions of CPQ$_{8-10}$ and CPQ$_{11-14}$. The Danish version of CPQ$_{8-10}$ had 27 questions (25 questions in four different domains: oral symptoms, functional limitations, social well-being and emotional well-being) and 2 global questions. The Danish version of CPQ$_{11-14}$ had 39 questions (37 questions in four different domains: oral symptoms, functional limitations, social well-being and emotional well-being) and 2 global questions. The
study population for CPQ8-10 consisted of 120 eight-to-10-year old children in three different groups: 1) healthy grade 4 children, 2) children with cleft lip and palate, and 3) children with rare oral conditions e.g. amelogenesis imperfecta, ectodermal dysplasia or tooth agenesis. For CPQ11-14, the study group included 225 eleven-to-14-year old children in four different groups: 1) healthy grade 6 children, 2) children with cleft lip and palate, 3) children with rare oral conditions and 4) children undergoing orthodontic treatment (within the first three months). For CPQ8-10, the mean overall CPQ score was significantly higher in children with rare oral conditions compared to the healthy children. The overall CPQ score was 15 for children with rare oral conditions, 7 for healthy children and 5 for children with cleft lip and palate. For CPQ11-14, the mean overall CPQ score was 22 for the orthodontic group, 17 for children with rare oral conditions, 9 for healthy children and 9 for children with cleft lip and palate. They showed that CPQ8-10 and CPQ11-14 are valid measures for assessing oral health-related quality of life in Danish children.

Humphris et al. (69) evaluated the validity and reliability of CPQ8-10 by means of confirmatory factor analysis (CFA) in Northern Ireland. They assessed oral health-related quality of life in a sample of 270 grade four children using CPQ8-10 and Coopersmith Self-Esteem Inventory-School Form (Coopersmith SEI-SF) for children aged 8-15. There was no statistically significant difference between overall CPQ score or self-esteem scale score and age. There was no statistically significant difference between overall CPQ score or self-esteem scale score and gender. The results of the study confirmed the validity and reliability of CPQ8-10.

Locker et al. (6) evaluated the oral health-related quality of life in children with oligodontia by means of CPQ 11-14. The study group consisted of a sample of 36 children with oligodontia aged 11-14 years who were recruited from the orthodontic clinic at Toronto’s Hospital for Sick
Children and Bloorview MacMillan Childrens Centre’s northern outreach cleft lip and palate/craniofacial dental clinics in the Ontario communities of Thunder Bay, Sudbury, Sault Ste. Marie, and Timmins. The study was based on convenient sample of children who presented to the above-mentioned clinics with no sample size calculation.

The questionnaire included 37 questions in four domains including oral symptoms, functional limitations, emotional well-being, and social well-being and two global rating questions. The sample studied had one to 14 missing permanent teeth with 69% of the sample having 6 or more missing teeth. The most common missing teeth were premolars, anterior teeth, and molars in descending order. They described some indicators such as “prevalence”, “extent” and “severity”. According to their definition, “prevalence” is the percentage of children experiencing one or more impacts “everyday/almost every day” or “often”. “Extent” of the impact is the mean number of items with a score of “everyday/almost every day” or “often”. “Severity” of the impact is the mean overall CPQ score.

The “prevalence” of overall CPQ score was 77.8%. The “prevalence” was 72.2% for oral symptoms, 77.8% for functional limitations, 41.7% for emotional well-being, and 58.3% for social well-being. The “extent” scores showed that functional limitations domain was the domain most affected (0.78) compared to other domains (oral symptoms: 0.28, emotional well-being: 0.42, social well-being: 0.28). The “severity” score was 22.3 for overall score, 5.0 for oral symptoms, 6.4 for functional limitations, 5.4 for emotional well-being and 5.7 for social well-being. There was no statistically significant difference between age or gender and the “prevalence”, “extent” or the “severity” of impacts. There was no significant correlation between total score or domain scores and the number of missing permanent teeth. There was no correlation between the mean CPQ score and the number of missing anterior teeth. The results
showed that oligodontia had a negative functional and psychological impact on patients. The findings of this study indicated that the OHRQoL of children with oligodontia is lower than children with dental caries or malocclusion but higher than children with orofacial conditions.

Wong et al. (60) used CPQ_{11-14} to assess oral health-related quality of life in a group of 25 patients aged 11-15 years with severe hypodontia (missing 4 or more permanent teeth excluding the third molars). The patients were recruited from pediatric dentistry and orthodontic clinics in Hong Kong. The number of missing teeth were reported in two ways: 1) number of missing permanent teeth, 2) number of missing permanent teeth accounting for retained primary teeth (They did not count the tooth as missing if there was a retained primary tooth in it’s place).

The number of permanent teeth missing in the maxillary arch was greater than in the mandibular arch by a ratio of 1.7 to 1. The mean number of missing permanent teeth was 8.9 (with the range of 4 to 20). The most commonly permanent missing tooth was the maxillary lateral incisor followed by the mandibular central incisor. Accounting for the retained primary teeth, the mean number of missing teeth was 3.7. The most commonly missing tooth after accounting for retained primary teeth was the maxillary lateral incisor followed by the mandibular central incisor by a ratio of 1.2 to 1. The mean overall CPQ score of 29 was reported. The mean oral symptoms score was 7.8, the mean functional limitations score was 6.5, the mean emotional well-being score was 8.4 and the mean social well-being score was 6.4. All participants experienced one or more impacts. There was a moderate correlation between total and domain scores and the number of missing permanent teeth (p < .05). Accounting for retained primary teeth, there was a strong correlation between the number of missing teeth and the total and domain scores (P < .001). The results of the study showed that severe hypodontia (missing four or more permanent teeth) has substantial impact on the oral health-related quality of life of the
affected children. It also emphasized the importance of retained primary teeth by suggesting that these teeth can mask the effect of severe hypodontia.

In 2006, Jokovic et al. (7) developed short formats of CPQ\textsubscript{11-14} using item impact method and stepwise regression method. The study group consisted of children in three categories: 1) pediatric dentistry group (children with dental caries), 2) orthodontic group, 3) orofacial group (children with cleft lip and palate). The short format questionnaire has some advantages over the long format questionnaire including being less time consuming, less costly for data collection, and having less chance for non-response. Instead of 37 questions in the original format, the short-form questionnaire consisted of two types: a 16-item version for use in clinical settings (with four items per domain) and an 8-item version for use in epidemiological studies (with two items per domain). There was a significant relationship between original CPQ\textsubscript{11-14} scores and short format scores. All short format questionnaires showed higher CPQ scores compared to the original CPQ\textsubscript{11-14}. The CPQ scores for short format questionnaires were reported in orofacial group, orthodontic group and pediatric dentistry group in descending order. The authors showed the validity and reliability of both short format questionnaires.

A study was performed by Foster Page et al. (8) in New Zealand to assess the reliability and validity of CPQ\textsubscript{8-10} and CPQ\textsubscript{11-14} in younger children. They assessed the oral health-related quality of life using the CPQ\textsubscript{8-10} (25 items) and short format of CPQ\textsubscript{11-14} (16-item impact) in 183 children who presented to dental clinics for dental treatment. The children were divided into two groups: younger children (5-to-6-year old) and older children (7-to-8-year old). The results of the study showed a strong positive correlation between the scores of CPQ\textsubscript{11-14} and CPQ\textsubscript{8-10} for all the children (P < .001). Both questionnaires showed higher scores in children with higher caries rate. Both questionnaires showed higher CPQ score in females compared to males. Children living in
“high deprivation” areas had higher CPQ score compared to children living in “low deprivation” areas. The study validated the use of CPQ_{11-14} for 5-8-year-old children and the authors suggested that the short format of CPQ_{11-14} can be used in 5-14-year-old children.

Laing et al. (70) used the CPQ_{11-14} to assess oral health-related quality of life in a group of 11-16-year-old children with hypodontia who were missing one or more permanent teeth (n=62) and a control group of routine orthodontic patients (n=61) in England. They used original format of CPQ_{11-14} with 37 questions and also two visual analogue scale questions (VAS) assessing appearance and function of the teeth. In order to account for retained primary teeth, the authors defined “relative” hypodontia (the difference between the number of missing permanent teeth and the number of retained primary teeth) versus “absolute” hypodontia (number of missing permanent teeth). The reported mean absolute hypodontia was 4.52, whereas mean relative hypodontia was 2.03 missing teeth in each subject. The mandibular middle sextant had the most missing teeth and maxillary middle sextant had the most relative hypodontia. The mean overall CPQ score for hypodontia group was 26.82 and for orthodontic group was 28.52. There was no significant relationship between the overall CPQ score in hypodontia group and age, gender, absolute hypodontia, and relative hypodontia. Functional limitations domain was significantly correlated with the relative hypodontia. Functional limitations domain scores were only affected by missing in the posterior maxillary and mandibular sextants. There was a significant relationship between relative hypodontia and VAS score for eating (VAS score increased as the relative hypodontia increased). There was no significant difference in overall CPQ score or domain scores between the two study groups.
2.7 Parental-Caregiver Perceptions Questionnaire (P-CPQ)

Despite the evidence supporting child self-reports on their oral health-related quality of life, parental perception is also vital as they are the main decision maker for their children’s treatment choices. Parental reports are considered as adjuncts to children’s report in assessing the quality of life in children.

Jokovic et al. (71) developed the Parental-Caregiver Perceptions Questionnaire (P-CPQ) and evaluated its validity and reliability. The questionnaire contained 31 questions with the response codes as never= 0, once or twice= 1, sometimes= 2, often= 3, every day or almost every day= 4, and “I don’t know” responses. Excluding “I don’t know” responses will lead to loss of information. In order to manage the “I don’t know” responses, the score was adjusted by giving the score of 0 to those responses and then calculating the CPQ score by dividing the sum of all 31 responses to the number of items with a valid response (i.e. reduce the denominator by the number of items having a response of “I don’t know.”) The mean overall P-CPQ was 25.1 (with the range of 0 to 80). The highest total score was reported in orofacial group (31.83), followed by the orthodontic group (23.84) and the pediatric dentistry group (5.86). The orofacial group also had highest domain scores for three domains including functional limitations, emotional well-being and social well-being domain. In the pediatric dentistry group, the P-CPQ score was higher for children with a higher caries rate. The results of the study supported the validity and reliability of P-CPQ.

In another study, the same authors (72) evaluated the level of agreement in OHRQoL between 42 pairs of 11-to-14-year old children with oral and orofacial conditions and their mothers using CPQ11-14 and P-CPQ, respectively. There was substantial agreement for overall scores (ICC: 0.70), excellent agreement for oral symptoms (ICC: 0.81), substantial agreement for
functional limitations (ICC: 0.71) and moderate agreement for social (ICC: 0.58) and emotional well-being (ICC: 0.52) between children and their mothers. There was moderate agreement for the global rating questions between mothers and their children. The level of agreement between mothers and their children varies based on some factors: lower level of agreement was noted in older children, females, orofacial group and children with highest scores for quality of life. The overall CPQ score reported by children (26.2) was higher than the score reported by their parents (22.6). The authors suggested that both children and their parents’ perception should be used in oral health-related quality of life assessment in children.

More recently, Kotecha et al. (9) evaluated OHRQoL in children by means of short format of CPQ11-14. Their study consisted of three groups: a group of 11-14-year-old children with mild tooth agenesis who were missing less than six teeth (n=43), a group with severe tooth agenesis who were missing six or more teeth (n=43) and a control group of children with orthodontic treatment needs (n=30). The Parental-Caregiver Perceptions Questionnaire (P-CPQ) was filled by parents of each participating child to compare OHRQoL perceived by parents to that of their children. The mean number of missing permanent teeth was 6.8 (with the range of 2-18). The most commonly missing permanent teeth were maxillary lateral incisors, mandibular second premolars and maxillary second premolars in descending order. The overall CPQ score and all domain scores were significantly higher in tooth agenesis group compared to the control orthodontic group. There was no significant correlation between the number of missing permanent teeth and OHRQoL in children. There was no significant difference in CPQ scores and age, gender, socioeconomic status, site of missing teeth (anterior versus posterior) and retained primary teeth. The correlation between child and parent OHRQol was found to be moderate (rho = 0.46, p < .001).
Ferreira et al. (73) used a Brazilian version of Parental-Caregiver Perceptions Questionnaire (P-CPQ) and the short format of CPQ11-14 to assess the agreement between 960 mothers and their 11-to-14-year old child’s perception regarding oral health-related quality of life. The total CPQ score reported by children was significantly higher than the CPQ score reported by mothers. The reported functional limitations score and social well-being score by children was signifyingly higher than their mothers. The results showed moderate agreement between children and their mothers for oral health-related quality of life.

In a study conducted by Kohli et al. (74), OHRQoL was assessed in a group of 35 children with hypohidrotic ectodermal dysplasia (HED) and their parents by means of CPQ11-14 and P-CPQ, respectively. The results showed that more functional limitations were experienced by older children (15-19 years old) compared to younger children (11-14 years old). There was no significant difference between children and their parents regarding total OHRQoL scores or domain scores. Female children with HED experienced more emotional limitations compared to male children.
2.8 Summary

Following a synthesis of the most relevant literature, it can be summarized that there is lack of clear understanding of the effect of oligodontia on the OHRQoL of the affected children. For a comprehensive understanding of the oral health-related quality of life, child reports need to be obtained. If a child’s cognitive level is sufficient to answer the questionnaire, child reports are preferable to parental reports. However, if the child is not able to provide information concerning their health status because of their age, a language barrier or developmental delay, parental reports should be used. Further research is required to assess the oral health-related quality of life based on both child and parental inputs in children suffering from oligodontia. Comparing the child input with parental input report is beneficial to developing a comprehensive treatment plan. It is also important to determine if other factors such as age, sex, and socioeconomic status impact the oral health-related quality of life in these patients.
2.9 Statement of the problem

The review of the literature showed that there are very few studies assessing OHRQoL in children with oligodontia. Most of the previous studies assessed OHRQoL in children with dental caries, cleft lip/palate or rare orofacial conditions but only a few studies on OHRQoL in children with oligodontia have been performed. There is a lack of a clear understanding on whether oligodontia affects OHRQoL in affected children and whether these impacts are perceived similarly by the affected children and their parents. Therefore, more research in this field is required to better understand the impacts of oligodontia on the OHRQoL in affected children.
Chapter 3

Aims and hypothesis

3.1 Aims

1. To assess oral health-related quality of life in children with oligodontia based on their self-report.

2. To determine the parents’ view of children with oligodontia regarding their children’s oral health-related quality of life.

3. To investigate the correlation between child and parents’ reported oral health-related quality of life in a sample of children with oligodontia.

4. To evaluate the effect of sex, age, socioeconomic status, number of missing permanent teeth, location of missing permanent teeth (maxillary, mandibular, anterior, middle, posterior) and retained primary teeth on children’s oral health-related quality of life.

3.2 Hypothesis

There is a strong correlation between the oral health-related quality of life reported by children affected by oligodontia and that reported by their parents.
Chapter 4

Materials and Methods

4.1 Study Population

This cross sectional cohort study was approved by the Health Sciences Research Ethics Boards at The Hospital for Sick Children and Holland Bloorview Kids Rehabilitation Hospital (Appendices 9-10).

Thirty-five 8- to 18-year-old patients who were diagnosed with oligodontia were recruited from dental clinics at The Hospital for Sick Children and Holland Bloorview Kids Rehabilitation Hospital between June 2017 and April 2018. In Toronto, patients with oligodontia are referred to both institutes to be assessed for their eligibility to be covered by the Cleft Lip and Palate/Craniofacial Dental (CLP/CD) Program of the Ontario Ministry of Health and Long-Term Care (MOHLTC). The patients included in this study were recruited either at their first visit to the hospital for an assessment of eligibility for the CLP/CD Program or at the time of assessment of prosthodontic treatment needs by a prosthodontist, but prior to starting any intervention for their oligodontia.

4.2 Inclusion Criteria

Patients were included in the study if they met the following criteria:

a) Scheduled for initial appointment for CLP/CD Program eligibility assessment or assessment appointment with the prosthodontist

b) Age range: 8 to 18 years old

c) Having at least 6 missing permanent teeth (excluding the third molars)

d) Patients and parents/legal guardians being able to read and write in English
4.3 Exclusion Criteria

Patients were not included if any of the following was true:

a) Under 8 or above 18 years of age

b) Less than 6 missing permanent teeth (excluding the third molars)

c) Patient or parent/legal guardian not being able to read or write in English

d) Having a systemic or developmental disorder or a known craniofacial anomaly or cleft of the lip and/or palate.

e) Having received any previous orthodontic, restorative or prosthodontic treatment intervention for oligodontia

4.4 Consent

The research investigator, the research coordinator at The Hospital for Sick Children or the clinical fellow at Holland Bloorview Kids Rehabilitation Hospital, at the respective sites, explained the study to eligible children and their parents/legal guardians. These investigators had been standardized to provide uniform instructions in a similar manner to prospective study participants.

The study was explained verbally to patients and their parents/legal guardians. It was described to the patients and their parents/legal guardians that separate study questionnaires were to be filled out by the patients and their parents/legal guardians and that patients would receive a clinical oral examination to record the missing and existing teeth. It was also discussed with the family that demographic data including age, sex and family income would be recorded.

It was explained to the families that their participation in the study was completely voluntary and if they declined to participate, it would not affect the care they would receive at the respective
hospitals. It was discussed with the patients and their parents that they could decline participation in the study at any time and for any reason during the study. It was also mentioned that data collected from the questionnaire was anonymized and all the data was confidential, being accessible only to the research team. If the patients and their parents wished to participate in the study, consent or assent form, whichever applicable, were obtained from the child. Parents’ or legal guardian’s consent was also obtained. The patient and their parents had the opportunity to ask questions and the research team explained anything that was not clear. One copy of the signed assent or consent form was given to the patient and their parent and one copy was kept by the research coordinator in a locked cabinet. A copy of these forms was also included in the patient’s health records. The consent and assent forms used in this study are included as Appendixes 4-8.

4.5 Procedure

Enrolled subjects received the appropriate questionnaires which were completed by them in their assigned clinic rooms. Both questionnaires were self-completed. Children completed the questionnaire on their own without parental or guardian help. Parents and children were asked not to discuss their questions and answers among themselves. In case of clarification needed by the children or the parents for any item in the questionnaire, the research investigator or the research coordinator helped to explain the question.

Children completed the short format of CPQ\textsubscript{11-14} (Appendix 1). This questionnaire was developed by Jokovic et al. in order to facilitate its use in clinical settings (7). We used the short format of CPQ\textsubscript{11-14} in our study based on the results of the study by Foster Page (8) who assessed the validity of short format of CPQ\textsubscript{11-14} in young children and suggested that this questionnaire can be used in children as young as 5 years old.
The short format of CPQ\textsubscript{11-14} consists of 16 questions in four domains:

a) Oral symptoms: history of pain, mouth ulcers, bad breath, food stuck between teeth

b) Functional limitations: longer feeding times, chewing problems, drinking problems, speech difficulty

c) Emotional well-being: irritable/frustrated, shy/embarrassed, concerned what other people think, upset

d) Social well-being: problems smiling/laughing, being teased, having arguments with family or other children, being asked questions regarding teeth, lips, jaws or mouth

Each patient answered the questions according to their experiences in the four different domains within the past three months. A score was assigned to each answer: never = 0, once/twice = 1, sometimes = 2, often =3, every day/almost every day=4. Response codes for each item were added to calculate the overall CPQ and domain scores.

Three single-item global ratings were also included in the short format of CPQ\textsubscript{11-14}: 1) asking the children to rate their oral health from “excellent” to “poor”, 2) how much their life was being affected by their oral condition from “not at all” to “very much”, 3) how satisfied the patients were with the appearance of their teeth ranging from “very satisfied” to “dissatisfied”.

One parent/guardian of each patient was asked to complete the P-CPQ (Appendix 2). This questionnaire had the same domains as the CPQ\textsubscript{11-14}. Response options were: never=0, once or twice=1, sometimes=2, often=3, every day or almost every day=4. All “don’t know” responses were given a value of 0.

The P-CPQ included two global ratings: 1) how the parents rated the oral health of their child from “excellent” to “poor”, 2) how much their child’s well-being had been affected by their oral
condition, from “not at all” to “very much”.

After the questionnaires were completed, all patients received a clinical dental examination which recorded the missing teeth, total number of missing permanent teeth, site of missing teeth (maxillary anterior, maxillary posterior, mandibular anterior, mandibular posterior), permanent teeth present and primary teeth present. Demographic data including age, sex and family income were also recorded (Appendix 3).

The missing teeth were confirmed by examining panoramic radiographs which were available as part of the patient records for CLP/CD Program assessment in the hospital for every patient included in this study. Patients were not exposed to any additional radiographic examination for the purpose of the study.

In order to account for retained primary teeth, we also recorded site-specific tooth absence. The edentulous areas of the dental arch where there were no primary or permanent teeth present were defined as site-specific tooth absence locations. If the patient had a retained primary tooth with missing permanent successor, we did not record that site as a site-specific tooth absence.

An oral hygiene kit of toothbrush, toothpaste and dental floss was given to each participating patient and the accompanying parent/guardian as described in the invitation for participating in the study to compensate for their time and effort in participating in this study.

**4.6 Data Collection and Management**

4.6.1 Storage of Patient Information

At both study sites, a study package consisting of CPQ11-14, P-CPQ and data collection forms was utilized for each patient who had enrolled in the study. Each study package had a study ID number with all personal health information or identifiers removed. We kept the completed
packages in a locked cabinet in a locked office. All these records were only accessible to the research investigator, research coordinator, principal investigator and co-investigators.

4.6.2 Data Entry and Storage

The study ID was used to enter patient’s data from the questionnaires and data collection forms in an anonymized manner at The Hospital for Sick Children and Holland Bloorview Kids Rehabilitation Hospital in order to protect patient confidentiality. This data was accessible only to the research investigator, research coordinator, principal investigator and co-investigators. The study continued until data was obtained from 35 children and 35 parents/legal guardians.

The following data were recorded (Appendix 3):

Study ID

Date of the examination

Demographic data: age, sex, family income, relationship of the parent (father, mother, legal guardian)

Missing permanent teeth

Total number of missing permanent teeth

Location of the missing permanent teeth: maxillary anterior, maxillary posterior, mandibular anterior, mandibular posterior

Erupted permanent teeth

Retained primary teeth

Site-specific tooth absence

Basic dental examination data: mixed dentition, permanent dentition
4.7 Statistical Analysis

Descriptive statistics were provided for demographic variables including age, gender and family income, for clinical variables including number of missing permanent teeth, number of site-specific tooth absences and location of missing permanent teeth, and for quality of life variables (i.e. children and parent CPQ scores). Means, standard deviations and ranges were used to describe continuous variables. Count and proportions were calculated for categorical variables. For CPQ scores, item responses in each CPQ domain were added to generate domain scores. The total score was calculated as the sum of the 4 domain scores. Children completed the short format of CPQ11-14 whereas parents completed the P-CPQ. The CPQ11-14 does not have any “I don’t know” responses whereas P-CPQ includes “I don’t know” in the responses. In order to compare children and parent responses, we computed mean score per item by using the total score divided by the number of items with valid responses. For parents, when calculating sum of item scores, “I don’t know” responses were given a value of 0. The number of items with valid responses was calculated by subtracting the number of “I don’t know” responses from the total number of questions (71). We examined the distribution and proportion of “I don’t know” responses in parents. The two-sample Wilcoxon test was used to compare the number of “I don’t know” responses in mothers and fathers. Paired t-tests were used to compare mean score per item between parents and children in regard to the total CPQ score and the domain scores. Univariate linear regression models were performed to examine the associations between CPQ score and variables of interest. Spearman’s correlation analyses were performed to examine the associations between children and parent scores in each domain and overall. Further, a multiple linear regression model was built to investigate the association between CPQ score and oligodontia (number of missing permanent teeth, site-specific tooth absence), while controlling
for confounders.

Sufficient sample size is required to detect estimated effect size. In order to account for correlations between parents and children from the same family, two-sided paired t-test was used in the sample size calculation. A sample size of 27 children and parent pairs achieved 81% power to detect a medium effect size of 0.5 at a significance level (alpha) of .05 using a two-sided paired t-test. The estimated mean of paired differences in adjusted scores (mean score per item accounting for number of “I don’t know” answers) was 0.30 and the estimated standard deviation of differences was 0.60. The sample size was calculated using PASS 12 (NCSS LLC, Kaysville, Utah), and all analyses were performed using SAS 9.4 (SAS Institute Inc., Cary, NC, USA).” In order to account for missing data, we recruited 35 patients and 35 parents.
Chapter 5

Results

A total of 35 children and 35 parents completed the CPQ\textsubscript{11-14} and P-CPQ, respectively. We approached 36 children and their families and only one family denied participation in the study because their child was unaware of her oligodontia and they did not want to inform the child. Therefore, the acceptance rate for our study was 97.2%. The average age of children was 12.36 years with the youngest being 8.17 years old and the oldest 17.58 years old. Among the 35 children, 10 (28.57\%) were male and 25 (71.43\%) were female. Sixteen (50.00\%) of the children belonged to the high-income group (yearly family income > $80,000) [Table 1].

<table>
<thead>
<tr>
<th>Variable</th>
<th>Mean ± SD or n (%)</th>
<th>Range</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (n=35)</td>
<td>12.36 ± 2.92</td>
<td>8.17-17.58</td>
<td></td>
</tr>
<tr>
<td>Gender - Male (n=35)</td>
<td>10 (28.57%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Number of missing permanent teeth (n=35)</td>
<td>9 ± 3</td>
<td>6 - 21</td>
<td>311</td>
</tr>
<tr>
<td>Site-specific tooth absence (n=35)</td>
<td>2 ± 3</td>
<td>0 - 11</td>
<td>64</td>
</tr>
<tr>
<td>Maxillary anterior (n=35)</td>
<td>26 (74.29%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Maxillary posterior (n=35)</td>
<td>34 (97.14%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mandibular anterior (n=35)</td>
<td>18 (51.43%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mandibular posterior (n=35)</td>
<td>32 (91.43%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Income groups</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>- 20000 - 40000</td>
<td>5 (15.63%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>- 40000 - 80000</td>
<td>11 (34.38%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>- &gt;80000</td>
<td>16 (50%)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Among the parents, there were 12 (36.36\%) fathers and 21 (63.64\%) mothers. Two parents did not indicate whether they were mothers or fathers.
Excluding the third molars (>82.86%), the most common missing permanent teeth were maxillary second premolars (68.57%), followed by maxillary lateral incisors (57.14%) and mandibular second premolars (57.14%). The most common missing permanent teeth within each quadrant were maxillary right second premolar (71.43%), maxillary left second premolar (65.71%), mandibular left second premolar (62.86%), maxillary left lateral incisor (57.14%), maxillary right lateral incisor (57.14%) and mandibular right second premolar (51.43%). The least commonly missing permanent teeth were mandibular left canine (0.00%), maxillary right central incisor (5.7%), maxillary left central incisor (5.71%), mandibular right canine (8.57%), mandibular left first molar (14.29%), mandibular right first molar (17.14%) and maxillary right first molar (17.14%) [Table 2].

In our sample studied, the missing of the third molars were seen in more than 82.86% of the subjects. Calcification of maxillary third molars and mandibular third molars occurs at 7-9 and 8-10 years of age, respectively (75). It is possible that in some of the subjects as they grow to an older age group, the third molars may develop, however third molars were not included in the consideration for definition of oligodontia in our study.
Table 2: Frequency and proportion of missing teeth

<table>
<thead>
<tr>
<th>Missing Permanent Teeth (FDI notation)</th>
<th>n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>18</td>
<td>34 (97.14%)</td>
</tr>
<tr>
<td>28</td>
<td>31 (88.57%)</td>
</tr>
<tr>
<td>38</td>
<td>30 (85.71%)</td>
</tr>
<tr>
<td>48</td>
<td>29 (82.86%)</td>
</tr>
<tr>
<td>15</td>
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<tr>
<td>25</td>
<td>23 (65.71%)</td>
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<tr>
<td>35</td>
<td>22 (62.86%)</td>
</tr>
<tr>
<td>22</td>
<td>20 (57.14%)</td>
</tr>
<tr>
<td>12</td>
<td>20 (57.14%)</td>
</tr>
<tr>
<td>45</td>
<td>18 (51.43%)</td>
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<tr>
<td>14</td>
<td>16 (45.71%)</td>
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<tr>
<td>47</td>
<td>14 (40.00%)</td>
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<tr>
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<td>11 (31.43%)</td>
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<td>11 (31.43%)</td>
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<tr>
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<td>11 (31.43%)</td>
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<td>10 (28.57%)</td>
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<tr>
<td>44</td>
<td>10 (28.57%)</td>
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<tr>
<td>23</td>
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<td>9 (25.71%)</td>
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<tr>
<td>31</td>
<td>9 (25.71%)</td>
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<tr>
<td>32</td>
<td>8 (22.86%)</td>
</tr>
<tr>
<td>42</td>
<td>8 (22.86%)</td>
</tr>
<tr>
<td>26</td>
<td>7 (20.00%)</td>
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<tr>
<td>16</td>
<td>6 (17.14%)</td>
</tr>
<tr>
<td>46</td>
<td>6 (17.14%)</td>
</tr>
<tr>
<td>36</td>
<td>5 (14.29%)</td>
</tr>
<tr>
<td>43</td>
<td>3 (8.57%)</td>
</tr>
<tr>
<td>11</td>
<td>2 (5.71%)</td>
</tr>
<tr>
<td>21</td>
<td>2 (5.71%)</td>
</tr>
<tr>
<td>33</td>
<td>0 (0%)</td>
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</tbody>
</table>
The CPQ_{11-14} was used to evaluate the quality of life in children. In global questions, 74.29% of children reported “good” to “excellent” oral health (Q1); however, 77.15% of children reported that their lives were affected by the condition of their teeth (Q2) and only 42.85% of children reported that they were satisfied with the appearance of their teeth (Q3) [Table 3].

<table>
<thead>
<tr>
<th>Global Questions</th>
<th>Count (proportions)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Child global Q1</strong></td>
<td></td>
</tr>
<tr>
<td>Excellent</td>
<td>1 (2.86%)</td>
</tr>
<tr>
<td>Very good</td>
<td>6 (17.14%)</td>
</tr>
<tr>
<td>Good</td>
<td>19 (54.29%)</td>
</tr>
<tr>
<td>Fair</td>
<td>9 (25.71%)</td>
</tr>
<tr>
<td>Poor</td>
<td>0 (0%)</td>
</tr>
<tr>
<td><strong>Child global Q2</strong></td>
<td></td>
</tr>
<tr>
<td>Not at all</td>
<td>8 (22.86%)</td>
</tr>
<tr>
<td>Very little</td>
<td>15 (42.86%)</td>
</tr>
<tr>
<td>Some</td>
<td>6 (17.14%)</td>
</tr>
<tr>
<td>A lot</td>
<td>5 (14.29%)</td>
</tr>
<tr>
<td>Very much</td>
<td>1 (2.86%)</td>
</tr>
<tr>
<td><strong>Child global Q3</strong></td>
<td></td>
</tr>
<tr>
<td>Very satisfied</td>
<td>2 (5.71%)</td>
</tr>
<tr>
<td>Satisfied</td>
<td>13 (37.14%)</td>
</tr>
<tr>
<td>Neither</td>
<td>12 (34.29%)</td>
</tr>
<tr>
<td>Dissatisfied</td>
<td>6 (17.14%)</td>
</tr>
<tr>
<td>Very dissatisfied</td>
<td>2 (5.71%)</td>
</tr>
</tbody>
</table>

*Q1: Asking the children to rate the health of their teeth, lips, jaws and mouth from “excellent” to “poor”.  
**Q2: How much does the condition of their teeth, lips, jaws and mouth affect their life overall from “Not at all” to “very much”.  
***Q3: How satisfied they are with the appearance of their teeth from “very satisfied” to “very dissatisfied”.
Parents completed the P-CPQ. For global questions, 68.57% of parents rated the health of their child’s oral condition “good” to “excellent” (Q1) and 80.01% of parents stated that their child’s overall well-being was affected by their oral condition (Q2) [Table 4].

Table 4: Global Questions in P-CPQ

<table>
<thead>
<tr>
<th>Total score</th>
<th>Count (proportions)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Parent global Q1*</td>
<td></td>
</tr>
<tr>
<td>Excellent</td>
<td>2 (5.71%)</td>
</tr>
<tr>
<td>Very good</td>
<td>11 (31.43%)</td>
</tr>
<tr>
<td>Good</td>
<td>11 (31.43%)</td>
</tr>
<tr>
<td>Fair</td>
<td>6 (17.14%)</td>
</tr>
<tr>
<td>Poor</td>
<td>5 (14.29%)</td>
</tr>
<tr>
<td>Parent global Q2**</td>
<td></td>
</tr>
<tr>
<td>Not at all</td>
<td>7 (20%)</td>
</tr>
<tr>
<td>Very little</td>
<td>11 (31.43%)</td>
</tr>
<tr>
<td>Some</td>
<td>12 (34.29%)</td>
</tr>
<tr>
<td>A lot</td>
<td>5 (14.29%)</td>
</tr>
<tr>
<td>Very much</td>
<td>0 (0.00%)</td>
</tr>
</tbody>
</table>

*Q1: How would the parents rate the health of their child’s teeth, lips, jaws and mouth from “excellent” to “poor”.

**Q2: How much their child’s overall well-being is affected by the condition of his/her teeth, lips, jaws or mouth from “not at all” to “very much”.
There were no “I don’t know” responses in children’s questionnaire, but parents’ questionnaire included “I don’t know” responses. For overall CPQ score, the average number of “I don’t know” responses reported by both mothers and fathers was 1.92 (p = .77) with 45.45% of parents reporting I don’t know responses.

For oral symptoms and functional limitations domains, the average number of “I don’t know” responses reported by fathers was 0.67 with 33.33% of fathers reporting “I don’t know” responses, which was not different from that reported by mothers (p = .91) with average number of 0.67 for “I don’t know” responses and 33.33% of mothers reporting “I don’t know” responses. For emotional well-being domain, 25.00% of fathers reported an average of 1.17 “I don’t know” responses, which was not different from the average of 1, reported by 28.57% of mothers (p = .87). For social well-being domain, 8.33% of fathers and 14.29% of mothers reported “I don’t know” responses. The average number of “I don’t know” responses was 0.08 for fathers and 0.24 for mothers with no difference between the two (p = .60).

According to the responses provided by subjects for each item, “prevalence” was calculated as the proportion of items with reported response of “often” or “every day/almost every day”. The “extent” was calculated as the mean number of items with response of “often” or “every day/almost every day”. “Severity” was defined as the mean number of items scored “often” or “every day/almost every day”. For children, prevalence in overall CPQ score, oral symptoms, functional limitations, emotional well-being and social well-being were as follows: 22 (62.86%), 8 (22.86%), 11 (31.42%), 11 (28.29%) and 6 (17.15%). The extent of impacts of oligodontia on each domain score and overall CPQ score were 1.5, 0.3, 0.4, 0.7 and 0.20, respectively. For parents, prevalence of items reported “often” or “every day/almost every day” in overall CPQ score, oral symptoms, functional limitations, emotional well-being and social well-being were 21
(61.87%), 15 (42.9%), 9 (25.71%) and 9 (26.5%), respectively. The corresponding mean numbers of items reported “often” or “every day/almost every day” were 1.9, 0.8, 0.6 and 0.5, respectively.

In children’s questionnaire, the average score for overall CPQ, oral symptoms, functional limitations, oral symptoms and functional limitations combined, emotional well-being and social well-being were 15.51, 4.46, 3.40, 7.86, 4.60 and 2.97, respectively. In parents’ questionnaire, the average score for overall CPQ, oral symptoms and functional limitations combined, emotional well-being, social well-being were 24.20, 8.63, 9.97 and 5.63, respectively.

When comparing mean score per item, paired-t tests suggested that children reported significantly higher response scores in overall CPQ (difference = 0.78 ± 0.51, p < .001), oral symptoms and functional limitations combined (difference = 0.82 ± 0.52, p < .001), emotional well-being (difference = 0.79 ± 0.91, p < .001), and social well-being (difference = 0.64 ± 0.65, p < .001). This suggested that children consistently reported poorer quality of life than parent perceptions, across all domains. In addition, Spearman’s correlation analyses were performed to examine the correlation between children and parent responses (mean score per item). No correlations were found in overall CPQ (ρ = 0.32, p = .06), oral symptoms and functional limitations combined (ρ = 0.05, p = .79) and social well-being (ρ = 0.32, p = .06). However, as children’s emotional well-being score increased, parental score also increased (ρ = 0.46, p = .006) [Table 5].
Univariate linear regression models were constructed to determine the associations between outcomes including overall CPQ and domain scores and predictors such as oligodontia (number of missing permanent teeth, site-specific tooth absence, location of missing permanent teeth) and patient demographics. Location of missing permanent teeth such as maxillary anterior (yes vs no) and mandibular anterior (yes vs no) were entered into the regression analyses. Since sparse cell counts could lead to unstable coefficient estimates, maxillary posterior (yes vs no) was not included because only 1 patient did not have missing permanent teeth at this location and mandibular posterior (yes vs no) was not included as only 3 patients did not have missing

<table>
<thead>
<tr>
<th>Table 5: Mean overall and domain scores in children and parents</th>
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<tbody>
<tr>
<td><strong>Total score</strong></td>
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<tr>
<td>Total score in Children</td>
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<tr>
<td>- Total score for oral symptoms (OS) in children</td>
</tr>
<tr>
<td>- Total score for functional limitation (FL) in children</td>
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<tr>
<td>- Total score for OS and FL in children</td>
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<tr>
<td>- Total score for emotional well-being (EW) in children</td>
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<tr>
<td>- Total score for social well-being (SW) in children</td>
</tr>
<tr>
<td>Total score in parents</td>
</tr>
<tr>
<td>- Total score for oral problems (OS) and functional limitation (FL) in parents</td>
</tr>
<tr>
<td>- Total score for emotional well-being (EW) in parents</td>
</tr>
<tr>
<td>- Total score for social well-being (SW) in parents</td>
</tr>
</tbody>
</table>
permanent teeth at this location. Similarly, mixed dentition and permanent dentition were not included in the univariate model because only 2 patients were in permanent dentition. Key variables of interests (number of missing permanent teeth and site-specific tooth absence) and factors showing trend or association in the univariate analyses were entered into the multiple regression models. Five multiple regression models were built using overall CPQ score and the four domain scores as the outcomes and the number of missing permanent teeth and site-specific tooth absence as predictors, while controlling for the effect of yearly family income (<80000 vs >80000) [Table 6].
Table 6: Multiple regression results

| Overall CPQ score as the outcome | Parameter                                      | Estimate | Standard Error | t Value | Pr > |t| |
|---------------------------------|------------------------------------------------|----------|----------------|---------|-------|---|
|                                 | Family income <80000 vs >80000                 | 5.03     | 3.03           | 1.66    | 0.11  |   |
|                                 | Number of missing permanent teeth              | 0.30     | 0.48           | 0.62    | 0.54  |   |
|                                 | Number of site-specific tooth absence          | 1.12     | 0.53           | 2.10    | 0.04  |   |
| Oral symptoms score as the outcome | Family income <80000 vs >80000                 | -0.11    | 0.84           | -0.13   | 0.90  |   |
|                                 | Number of missing permanent teeth              | 0.007    | 0.13           | 0.05    | 0.96  |   |
|                                 | Number of site-specific tooth absence          | 0.07     | 0.15           | 0.48    | 0.64  |   |
| Functional limitations score as the outcome | Family income <80000 vs >80000                 | 0.70     | 1.05           | 0.66    | 0.51  |   |
|                                 | Number of missing permanent teeth              | 0.25     | 0.17           | 1.51    | 0.14  |   |
|                                 | Number of site-specific tooth absence          | 0.28     | 0.19           | 1.49    | 0.15  |   |
| Emotional well-being score as the outcome | Family income <80000 vs >80000                 | 2.78     | 1.68           | 1.65    | 0.11  |   |
|                                 | Number of missing permanent teeth              | -0.02    | 0.27           | -0.07   | 0.94  |   |
|                                 | Number of site-specific tooth absence          | 0.44     | 0.30           | 1.49    | 0.15  |   |
| Social well-being score as the outcome | Family income <80000 vs >80000                 | 1.79     | 0.87           | 2.06    | 0.05  |   |
|                                 | Number of missing permanent teeth              | 0.009    | 0.14           | 0.06    | 0.95  |   |
|                                 | Number of site-specific tooth absence          | 0.36     | 0.15           | 2.37    | 0.03  |   |
Using overall CPQ score as the outcome, univariate results suggested that only the number of site-specific tooth absence was significant ($p = .02$). Age ($p = .26$), gender ($p = .90$), family income ($p = .08$), number of missing permanent teeth ($p = .40$), maxillary anterior missing ($p = .20$) and mandibular anterior missing ($p = .25$) were not related to overall score. Multiple regression model analyses revealed that as the number of site-specific tooth absence increased by 1, the overall CPQ score increased by 1.12 ($\beta = 1.12, \text{SE} = 0.53, p = .04$) (Figure 1). However, the overall CPQ scores were not correlated with the number of missing permanent teeth ($p = .54$) (Figure 2). Family income ($p = .11$) also remained statistically not significant. Using social well-being as the outcome, multiple regression results suggested that children from high-income family (>80000) reported significantly lower social well-being score than those from other income groups and the average difference in well-being score was 1.79 ($\beta = 1.79, \text{SE} = 0.87, p = .05$). Also, as the number of site-specific tooth absence increased by 1, social well-being score increased by 0.36 ($\beta = 0.36, \text{SE} = 0.15, p = .03$). No significant associations were found using oral symptoms, functional limitations or emotional well-being as the outcomes. Normality assumptions of residuals in these regression models were checked and met.
Figure 1: Overall CPQ scores in relation to number of site-specific tooth absences.

Figure 2: Overall CPQ scores in relation to number of missing permanent teeth.
Chapter 6
Discussion

There have been a considerable number of studies on the prevalence, etiology and treatment approaches of oligodontia but limited studies on the effects of oligodontia on the Oral Health-Related Quality of Life (OHRQoL), especially in children. Previously, parents have been questioned as the source of information for assessment of quality of life of their children but nowadays, with valid and reliable questionnaires becoming available, information can be obtained from children regarding their (OHRQoL) and the parent’s view can be considered as an adjunct to the child’s view, not a substitute.

This prospective cohort study was performed in order to assess OHRQoL in 35 pairs of 8-to-18-year-old children with oligodontia and their parents/legal guardians. The average age of children was 12.36 years (range: 8.17-17.58). The majority of the children in our sample were females (71.43%) which was similar to some studies (6,9,74), but in contrast to some others in which the majority of the subjects in the hypodontia group were males (60,70).

In this study, the most common missing permanent teeth other than the third molars were maxillary second premolars, followed by maxillary lateral incisors and mandibular second premolars (Table 1). There has been an inconsistency in the literature reporting the most prevalent missing permanent teeth. According to the study by Celikoglu et al. (4) the most common missing permanent teeth in the Turkish population were maxillary lateral incisors (46.5%), followed by mandibular second premolars (22.5%) and mandibular central incisors (13.4%). Similarly, in a study by Amini et al. the most common missing permanent teeth in the Iranian population were maxillary lateral incisors (37.2%), mandibular second premolars (22.1%), and mandibular central incisors (10.7%) (2). In the study by Sisman et al. (11) the most
common missing permanent teeth in the Turkish population were maxillary lateral incisors, followed by mandibular second premolars and maxillary second premolars. Gupta et al. (3) reported the most common missing permanent teeth in the Indian population were maxillary lateral incisors (1.69%), followed by mandibular central incisors (0.62%), maxillary premolars (0.53%), maxillary central incisors (0.53%) and mandibular premolars (0.45%). In a meta-analysis by Mattheeuws et al. (24), the most common missing permanent teeth were maxillary lateral incisors, mandibular second premolars, maxillary second premolars, and maxillary first premolars. The types of missing permanent teeth in our study are similar to those reported in the above-mentioned studies but with different frequencies.

We used the short format of CPQ11-14 to assess OHRQoL in children. This questionnaire contains three global rating questions. In global questions, 74.29% of children reported “good” to “excellent” oral health (Q1); however, 77.15% of children reported that their lives were affected by the condition of their teeth from “very little” to “very much” (Q2). 42.85% of children reported that they were “very satisfied” or “satisfied” with the appearance of their teeth (Q3) (Table 2). These findings are consistent with the study by Locker et al. which used the original format of CPQ11-14 in assessing OHRQoL in children with oligodontia. Their questionnaire had two global questions and their results showed that 63.9% of children reported “good” to “excellent” oral health (Q1), and 41.7% of children reported that their lives were being affected by their oral condition from “some” to “very much” (6).

In the present study, parents completed P-CPQ which had two global rating questions. 68.57% of the parents reported their children’s oral health as “good” to “excellent” (Q1), and 80.01% of parents reported that their child’s overall well-being has been affected by their oral condition from “very little” to “a lot”. There are no reports of the global rating questions in P-CPQ in the
previous studies. Unlike CPQ11-14, the P-CPQ contains “I don’t know” responses. In total, 45.45% of parents reported “I don’t know” responses in P-CPQ (15/33 parents).

The majority of parents in our study were mothers (63.64%). This is similar to studies by Jokovic (71) and Kotecha (9). There was no significant difference in the number of “I don’t know” responses between mothers and fathers for the overall CPQ score or for the domain scores. Similarly, in the study by Kotecha et al. which assessed OHRQoL in children with tooth agenesis and their parents using short format of CPQ11-14 and P-CPQ, respectively (9). Their results showed that there was no significant difference between mothers and fathers reports of their child’s quality of life. Therefore, both parents can provide valuable information regarding their children’s quality of life.

The frequency of “often” or “every day/almost every day” items (prevalence) reported by children were as follows: overall CPQ score: 62.86%, oral symptoms: 22.86%, functional limitations: 31.4%, emotional well-being: 31.42%, social well-being: 17.15%. The mean number of items with the score of “often” or “every day/almost every day” (the extent) was 1.5 for overall CPQ, 0.3 for oral symptoms, 0.4 for functional limitations, 0.7 for emotional well-being and 0.2 for social well-being. These findings were in agreement with the study by Kotecha et al. in the United Kingdom (9). In their study, the reported prevalence was 29% for oral symptoms, 30% for functional limitations, 33% for emotional well-being and 29% for social well-being. On the other hand, the study by Locker et al. (6) showed that the frequency of overall CPQ score was 77.8%, 27.8% for oral symptoms, 19.4% for emotional well-being and 16.7% for social well-being. The reported frequency of functional limitations in the study by Locker et al. was higher (61.1%) compared to the results of our study (31.4%). The extent (the mean number of items with the score of “often” or “every day/almost every day”) reported by Locker was in
agreement with the results of our study for overall score (1.75), oral symptoms (0.28), emotional well-being (0.42) and social well-being (0.28). However, for functional limitations, they reported a higher score (0.78). This difference can be due to having more retained primary teeth in the younger participants of our study as the subjects may not have been significantly affected by oligodontia due to the presence of the primary teeth. In the study by Wong et al. (60) which assessed OHRQoL in children with hypodontia using CPQ11-14, 100% of the participants reported experiencing oral symptoms, 88% having functional limitations, 88% having emotional well-being and 100% experiencing social well-being. One can speculate that the difference may be due to the threshold criteria used. For the prevalence, we reported the items with the response of “often” or “every day/almost every day” (similar to the criteria used by Locker et al. (6) and Kotecha et al.(9)). While it can be understood that if Wong et al. may have included “sometimes” responses in their criteria, then the reported prevalence would be expected to be higher.

The average overall CPQ score for children in our study was 15.51, 4.46 for oral symptoms, 3.40 for functional limitations, 4.60 for emotional well-being, and 2.97 for social well-being. The overall CPQ score in our study was lower compared to the studies by Wong et al. (29.0) (60), Laing (26.8) (70), and Locker et al. (22.3) (6). However, in the Kotecha et al. study (9), the overall CPQ score (16.0) was in agreement with our study. The domain scores in our study were lower compared to the scores reported by Locker et al. (6): oral symptoms: 5, functional limitations: 6.4, emotional well-being: 5.4 and social well-being: 3.0. The domain scores in the study by Wong et al. (60) were also higher than our study: oral symptoms: 7.8, functional limitations: 6.5, emotional well-being: 8.4 and social well-being: 6.4. The domain scores in our study was similar to those of Kotecha et al. study (9): oral symptoms: 4, functional limitations: 3,
emotional well-being: 5 and social well-being: 3. The lower CPQ overall score and domain scores compared to some of the above mentioned studies can be due to the fact that our study included younger patients compared to the previous studies, and consequently these subjects have higher number of retained primary teeth compared to older children.

The frequency of “often” or “every day/almost every day” responses reported by the parents was 61.87% for overall score, 42.85% for oral symptoms and functional limitations, 25.73% for emotional well-being and 26.47 for social well-being. The mean number of items reported by the parents “often” or “every day/almost every day” was 1.9 for the overall score, 0.8 for the oral symptoms and functional well-being, 0.6 for emotional well-being and 0.5 for the social well-being. The prevalence of these items has not been reported in the previous similar studies.

The parent’s reported overall CPQ score was 24.20. It was 8.63 for oral symptoms and functional limitations combined, 9.97 for emotional well-being, and 5.63 for social well-being. Children reported poorer quality of life compared to what was reported by their parents: children had significantly higher overall CPQ scores and domain scores including oral symptoms and functional limitations combined, emotional well-being and social well-being scores (p < .0001). This was similar to the findings by Ferreira et al. (73) and Jokovic et al. (6) who reported higher CPQ scores and domain scores in children compared to their parents. In the study by Kohli et al. that compared OHRQoL in children with ectodermal dysplasia and their parents (74), there was no significant difference in the parents reported overall CPQ score and domain scores compared to the scores reported by children.

The correlations between children and parents in overall CPQ score (ρ = 0.32, p = .06), oral symptoms and functional limitations (ρ = 0.05, p = .79) and social wellbeing (ρ = 0.32, p = .06) were not statistically significant. However, as children’s emotional wellbeing score increased,
parental score also increased ($\rho = 0.46, p = .006$). This is in contrast to the study by Kohli et al. (74) which reported a significant positive correlation for overall CPQ score and oral symptoms, functional limitations and emotional well-being but no significant correlation for the social well-being domain. In the study by Kotecha et al. (9), there was moderate correlation between children and their parents reported quality of life. Our study included younger patients compared to the above-mentioned studies, therefore the differences could be the result of age difference.

The overall CPQ scores were not correlated with the number of missing permanent teeth ($p = .54$). This is similar to the findings by Kotecha et al. (9). We hypothesize that this is due to the presence of primary teeth in the sites of permanent tooth agenesis, which was seen in 45.71% of the affected individuals, which could have potentially reduced the severity of impact on the OHRQoL and explain this lack of correlation with the overall CPQ. This explanation is further reinforced by the number of site-specific tooth absence that was significantly correlated with the overall CPQ scores ($p = .02$). Similarly, in the study by Laing et al. (70), there was no significant relationship between CPQ scores and absolute hypodontia (number of missing permanent teeth) or relative hypodontia (number of missing permanent teeth minus the number of retained primary teeth). This is in contrast to the findings by Kotecha et al. (9) who did not find any relationship between retained primary teeth and CPQ scores. It is worth mentioning that in their study, they only assessed the presence or absence of retained primary teeth as “yes” or “no”, therefore their results are not directly comparable with ours. In our study, we calculated site-specific tooth absence as the edentulous area with missing permanent tooth with no retained primary tooth in that area. In the study by Wong et al. (60), there was a moderate correlation between CPQ scores and the number of missing permanent teeth but there was a strong correlation between CPQ scores and the number of missing permanent teeth accounting for
retained primary teeth. These explanations describe that having a greater number of retained primary teeth in our sample could have masked the effect of hypodontia on the OHRQoL.

There was no correlation between the overall CPQ scores and location of tooth agenesis e.g. missing permanent in the anterior maxilla (p = .20) or missing permanent in the anterior mandible (p = .25). This is similar to the results of the study by Laing et al. (70) who did not find any significant relationship between missing permanent teeth in the maxillary middle sextant and the overall CPQ score. Kotecha et al. (9) found that patients with missing permanent teeth in the anterior segment had significantly higher social well-being score compared to patients with missing permanent teeth in the posterior segment. In our study, having retained primary teeth could have masked the effect of missing permanent teeth in different locations as explained previously.

There was no correlation between the overall CPQ scores and age (p = .26) and gender (p = .90). This is in agreement with the previous studies in this field by Locker et al. (6), Kotecha et al. (9) and Laing et al. (70). These results describe that gender and age of the child may have lesser impacts on the effects of oligodontia on OHRQoL.

There was no correlation between the overall CPQ score and family income (p = .08). However, high-income families (>80000 $) had significantly lower social well-being score than those from other income groups (p = .05). This is in agreement with the study by Kotecha et al. (9) who did not find any statistically significant difference between CPQ score and socioeconomic status using social deprivation score.

To our knowledge, this is the second study assessing OHRQoL in children with oligodontia based on their self-report and their parent’s report. The first reported study was performed by
Kotecha et al.(9) in the United Kingdom. We included the younger age group in our study because the effects of oligodontia on 8-to-10-year-old children have not been assessed in the previous studies. Children are usually diagnosed with oligodontia at early mixed dentition and are then referred for the assessment by the multidisciplinary treatment approach.

Strengths and limitations of this study should be considered. The published literature describing measurable impacts of oligodontia on OHRQoL in children is sparse and therefore the findings from a contemporary Canadian population are both novel as well as valuable in terms of understanding these impacts and providing perspective and rationale for planning publicly funded programs such as the province of Ontario’s Ministry of Health and Long-Term Care Cleft Lip and Palate/Craniofacial Dental program. At the same time, while these findings are important, it is equally important to recognize that this was a cross-sectional study based on a small sample size from hospital clinics. Its findings are, therefore, to be interpreted with caution. The sample size of the current study was sufficient for detecting overall CPQ score differences as low as 0.5 between children with oligodontia and their parents. However, this sample may be limited in representing the population as recruitment into the study depended upon referral of the affected children to the hospital for eligibility for funding through the aforementioned program. Therefore, the sample could have been affected by referral bias. We excluded patients who could not read/write in English and this could have excluded valuable information about the impacts of oligodontia on OHRQoL that could have been obtained from non-English speaking families. Patients and parents were asked to fill the questionnaires according to their experiences during the past three months which may have been affected by recall bias.

Clinical implications of this study are exemplified by the finding that children may be having impacts of oligodontia that may not be as well understood by their parents, therefore it is
important to consider these impacts when treating a child with oligodontia and to inform the parents so that these impacts can be offset to the extent possible at home in addition to whatever is needed to be done in the clinic setting through restorative treatment of these children.

**Future Directions**

Future studies could build upon the findings of the current investigation and further explore the directions this study has revealed. Future investigations could be planned to study older children in order to elicit the effects of oligodontia as these children grow to an age where some or many of their retained primary teeth may have exfoliated and the impacts on Oral Health-Related Quality of Life when these sites become edentate may become more pronounced. Longitudinal cohort studies on patients with oligodontia in the age groups where the orthodontic, restorative and prosthodontic phase of treatment has been completed can be planned to understand the longitudinal effects of the treatment of oligodontia. This will help to inform federal and provincial funding programs to better understand and invest in such treatments.
Chapter 7

Conclusions

In the sample studied in this cross-sectional cohort study, impacts of oligodontia on oral health-related quality of life in children were reported by both affected children and their parents.

1. Children’s overall CPQ score and domain scores were significantly higher than the scores reported by their parents indicating that children had poorer OHRQoL compared to what was perceived by their parents.

2. Children’s and parents’ reported overall CPQ score, oral symptoms and functional limitations, and social well-being were not correlated. However, as children’s emotional well-being score increased, parental score also increased.

3. There was a significant correlation between overall CPQ score reported by affected children and site-specific tooth absence.

4. Impacts of oligodontia on children or their parents were not related to the number of missing permanent teeth or the location of their missing permanent teeth.

5. There was no significant effect of age and gender on the OHRQoL of children with oligodontia.
References


21. Rølling S, Poulsen S. Short Communication Oligodontia in Danish schoolchildren. Acta


40. Gurrieri F, Franco B, Toriello H, G N. Oral-facial-digital syndromes: review and


59. Anweigi L, Allen PF, Ziada H. The use of the oral health impact profile to measure the


Appendix 1: Child Perception Questionnaire

Child Perceptions Questionnaire

Todays Date 

Participant ID: 

CHILD ORAL HEALTH QUESTIONNAIRE

Hello,

Thanks for agreeing to help us with our study!

This study is being done so that there will be more understanding about problems children may have because of their teeth, mouth, lips and jaws. By answering the questions, you will help us learn more about young people's experiences.

PLEASE REMEMBER:

• Don't write your name on the questionnaire
• This is not a test and there are no right or wrong answers
• Answer as honestly as you can. Don't talk to anyone about the questions when you are answering them. Your answers are private; no one you know will see them
• Read each question carefully and think about your experiences in the past 3 months when you answer
• Before you answer, ask yourself: "Does this happen to me because of problems with my teeth, mouth, lips and jaws?"
• Put an ☒ in the box for the answer that is best for you
FIRST, A FEW QUESTIONS ABOUT YOU

1. Would you say the **health** of your teeth, lips, jaws and mouth is:
   - [ ] Excellent
   - [ ] Very good
   - [ ] Good
   - [ ] Fair
   - [ ] Poor

2. How much does the condition of your teeth, lips, jaws or mouth affect your life overall?
   - [ ] Not at all
   - [ ] Very little
   - [ ] Some
   - [ ] A lot
   - [ ] Very much

3. How satisfied are you with the appearance of your teeth?
   - [ ] Very satisfied
   - [ ] Satisfied
   - [ ] Neither satisfied or dissatisfied
   - [ ] Dissatisfied
   - [ ] Very dissatisfied
QUESTIONS ABOUT ORAL PROBLEMS

In the past 3 months, how often have you had:

4. Pain in your teeth, lips, jaws or mouth?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day

5. Sores in your mouth?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day

6. Bad breath?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day

7. Food stuck in or between your teeth?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day
For the next question...

Has this happened because of your teeth, mouth, lips and jaws?

In the past 3 months, how often have you:

8. Taken longer than others to eat a meal?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day

In the past 3 months, because of your teeth, mouth, lips and jaws, how often has it been:

9. Difficult to bite or chew food like apples, corn on the cob or steak?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day

10. Difficult to say any words?
    - Never
    - Once or twice
    - Sometimes
    - Often
    - Everyday or almost every day
11. **Difficult to drink or eat hot or cold foods?**

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost every day

**QUESTIONS ABOUT FEELINGS**

**Have you had the feeling because of your teeth, mouth, lips and jaws?**
If you felt this way for another reason, answer 'Never'.

**In the past 3 months, how often have you:**

12. **Felt irritable or frustrated?**

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost every day

13. **Felt shy or embarrassed?**

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost every day
In the past 3 months, because of your teeth, mouth, lips and jaws, how often have you:

14. Been concerned what other people think about your teeth, mouth, lips and jaws?
   - □ Never
   - □ Once or twice
   - □ Sometimes
   - □ Often
   - □ Everyday or almost every day

15. Worried that you are not as good-looking as others?
   - □ Never
   - □ Once or twice
   - □ Sometimes
   - □ Often
   - □ Everyday or almost every day

16. Been upset?
   - □ Never
   - □ Once or twice
   - □ Sometimes
   - □ Often
   - □ Everyday or almost every day
QUESTIONS ABOUT YOUR SPARE-TIME ACTIVITIES & BEING WITH OTHER PEOPLE

Have you had these experiences because of your teeth, mouth, lips and jaws? If it was for another reason, answer 'Never'.

In the past 3 months, how often have you:

17. Avoided smiling or laughing when around other children?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day

18. Argued with other children or your family?
   - Never
   - Once or twice
   - Sometimes
   - Often
   - Everyday or almost every day
In the past 3 months, because of your teeth, mouth, lips and jaws, how often have:

19. Other children teased you or called you names?
   - [ ] Never
   - [ ] Once or twice
   - [ ] Sometimes
   - [ ] Often
   - [ ] Everyday or almost every day

20. Other children asked you questions about your teeth, lips, jaws or mouth?
   - [ ] Never
   - [ ] Once or twice
   - [ ] Sometimes
   - [ ] Often
   - [ ] Everyday or almost every day
THERE, IT'S FINISHED!

We appreciate the time and thought you have given to this questionnaire

THANK YOU FOR HELPING US

Derived by
Community Dental Health Services Research Unit
Faculty of Dentistry, University of Toronto
124 Edward Street, Toronto ON, M5G 1G6

Supported by: The Hospital for Sick Children Foundation
Appendix 2: Parental-Caregiver Perception Questionnaire

Parental-Caregiver Perceptions Questionnaire

Participant ID: ____________

COMMUNITY DENTAL HEALTH SERVICES RESEARCH
UNIT FACULTY OF DENTISTRY UNIVERSITY OF TORONTO
124 Edward Street Toronto, Ontario
M5G 1G6

CHILD ORAL HEALTH QUESTIONNAIRE
Parental report
6-14 years
INSTRUCTIONS TO PARENTS

1. This questionnaire is about the effects of oral conditions on children's well-being and everyday life, and the effects on their families. We are interested in any condition that involves teeth, lips, mouth or jaws. Please answer each question.

2. To answer the question please put an ☐ in the box by the response.

3. Please give the response that best describes your child's experience. If the question does not apply to your child, please answer with "Never".

Example: How often has your child had a hard time paying attention in school?

If your child has had a hard time paying attention in school because of problems with his/her teeth, lips, mouth or jaws, choose the appropriate response. If it has happened for other reasons, choose "Never".

☐ ☐ ☐ ☐ ☐ ☐
Never Once or twice Sometimes Often Everyday or Don't know almost everyday

4. Please do not discuss the questions with your child, as we are interested only in the parents' perspective in this questionnaire.
SECTION 1: Child's oral health and wellbeing

1. How would you rate the health of your child's teeth, lips, jaws and mouth?
   □ □ □ □ □
   Excellent Very good Good Fair Poor

2. How much is your child's overall wellbeing affected by the condition of his/her teeth, lips, jaws or mouth?
   □ □ □ □ □
   Not at all Very little Some A lot Very much

SECTION 2: The following questions ask about symptoms and discomfort that children may experience due to the condition of their teeth, lips, mouth and jaws

During the last 3 months, how often has your child had:

3. Pain in the teeth, lips, jaws or mouth?
   □ □ □ □ □ □
   Never Once or twice Sometimes Often Everyday or almost everyday Don't know
4. Bleeding gums?

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost everyday
- [ ] Don’t know

5. Sores in the mouth?

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost everyday
- [ ] Don’t know

6. Bad breath?

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost everyday
- [ ] Don’t know

7. Food stuck in the roof of the mouth?

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost everyday
- [ ] Don’t know

8. Food caught in or between the teeth?

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost everyday
- [ ] Don’t know

9. Difficulty biting or chewing foods such as fresh apple, corn on the cob or firm meat?

- [ ] Never
- [ ] Once or twice
- [ ] Sometimes
- [ ] Often
- [ ] Everyday or almost everyday
- [ ] Don’t know
During the last 3 months, because of his/her teeth, lips, mouth, or jaws, how often has your child:

<table>
<thead>
<tr>
<th>Question</th>
<th>Never</th>
<th>Once or twice</th>
<th>Sometimes</th>
<th>Often</th>
<th>Everyday or almost everyday</th>
<th>Don’t know</th>
</tr>
</thead>
<tbody>
<tr>
<td>10. Breathed through the mouth?</td>
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<tr>
<td>11. Had trouble sleeping?</td>
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<td></td>
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<tr>
<td>12. Had difficulty saying any words?</td>
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<tr>
<td>13. Taken longer than others to eat a meal?</td>
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<tr>
<td>14. Had difficulty drinking or eating hot or cold foods?</td>
<td></td>
<td></td>
<td></td>
<td></td>
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</tr>
<tr>
<td>15. Had difficulty eating foods he/she would like to eat?</td>
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</tr>
</tbody>
</table>
16. Had diet restricted to certain types of food (e.g. soft food)?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday

17. Upset?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday

18. Irritable or frustrated?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday

19. Anxious or fearful?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday

SECTION 3: The following questions ask about the effects that the condition of children’s teeth, lips, mouth and jaws may have on their feelings and everyday activities:

During the last 3 months, because of his/her teeth, lips, mouth or jaws, how often has your child been:
During the last 3 months, because of his/her teeth, lips, mouth or jaws, how often has your child:

20. Missed school (e.g. pain, appointments, surgery)?

   □ □ □ □ □
   Never Once or twice Sometimes Often Everyday or almost everyday Don't know

21. Had a hard time paying attention in school?

   □ □ □ □ □
   Never Once or twice Sometimes Often Everyday or almost everyday Don't know

22. Not wanted to speak or read out loud in class?

   □ □ □ □ □
   Never Once or twice Sometimes Often Everyday or almost everyday Don't know

23. Not wanted to talk to other children?

   □ □ □ □ □
   Never Once or twice Sometimes Often Everyday or almost everyday Don't know

24. Avoided smiling or laughing when around other children?

   □ □ □ □ □
   Never Once or twice Sometimes Often Everyday or almost everyday Don’t know
During the last 3 months, because of his/her teeth, lips, mouth or jaws, how often has your child:

<table>
<thead>
<tr>
<th>Question</th>
<th>Never</th>
<th>Once or twice</th>
<th>Sometimes</th>
<th>Often</th>
<th>Everyday or almost everyday</th>
<th>Don’t know</th>
</tr>
</thead>
<tbody>
<tr>
<td>25. Worried that he/she is not as healthy as other people?</td>
<td></td>
<td></td>
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<tr>
<td>26. Worried that he/she is different than other people?</td>
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</tr>
<tr>
<td>27. Worried that he/she is not as good-looking as other people?</td>
<td></td>
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<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>28. Acted shy or embarrassed?</td>
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</tr>
<tr>
<td>29. Been teased or called names by other children?</td>
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<td></td>
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<td></td>
<td></td>
</tr>
<tr>
<td>30. Been left out by other children?</td>
<td></td>
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<td></td>
<td></td>
</tr>
</tbody>
</table>
31. Not wanted or been unable to spend time with other children?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday  ☐ Don’t know

32. Not wanted or been unable to participate in activities such as sports, clubs, drama, music, school trips?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday  ☐ Don’t know

33. Worried that he/she has fewer friends?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday  ☐ Don’t know

**During the last 3 months, how often has your child been:**

34. Concerned what other people think about his/her teeth, lips, mouth or jaws?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday  ☐ Don’t know

35. Asked questions by other children about his/her teeth, lips, mouth or jaws?

☐ Never  ☐ Once or twice  ☐ Sometimes  ☐ Often  ☐ Everyday or almost everyday  ☐ Don’t know

9
SECTION 4: The following questions ask about effects that a child’s oral condition may have on PARENTS AND OTHER FAMILY MEMBERS

During the last 3 months, because of your child’s teeth, lips, mouth or jaws, how often have you or another family member:

36. Been upset?

☐ Never ☐ Once or twice ☐ Sometimes ☐ Often ☐ Everyday or almost everyday ☐ Don’t know

37. Had sleep disrupted?

☐ Never ☐ Once or twice ☐ Sometimes ☐ Often ☐ Everyday or almost everyday ☐ Don’t know

38. Felt guilty?

☐ Never ☐ Once or twice ☐ Sometimes ☐ Often ☐ Everyday or almost everyday ☐ Don’t know

39. Taken time off work (e.g. pain, appointments, surgery)?

☐ Never ☐ Once or twice ☐ Sometimes ☐ Often ☐ Everyday or almost everyday ☐ Don’t know
40. Had less time for yourself or the family?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

41. Worried that your child will have fewer life opportunities (e.g. for dating, getting married, having children, getting a job he/she will like)?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

42. Felt uncomfortable in public places (e.g. stores, restaurants) with your child?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

During the last 3 months, because of his/her teeth, lips, mouth, or jaws, how often has your child:

43. Been jealous of you or others in the family?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

44. Blamed you or another person in the family?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know
45. Argued with you or others in the family?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

46. Required more attention from you or others in the family?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

During the last 3 months, how often has the condition of your child’s teeth, lips, mouth or jaws?

47. Interfered with family activities at home or elsewhere?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

48. Caused disagreement or conflict in your family?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know

49. Caused financial difficulties for your family?

Never  Once or twice  Sometimes  Often  Everyday or almost everyday  Don’t know
SECTION 5: Child's gender and age:

a. Your child is:
   ☐ MALE
   ☐ FEMALE

b. Your child's age is: _____ YEARS

Questionnaire completed by:
   ☐ MOTHER
   ☐ FATHER
   ☐ OTHER

Date completed: ___/___/_____
Appendix 3: Data Collection Form

### Demographic Data

<table>
<thead>
<tr>
<th>Data</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Study ID</td>
<td></td>
</tr>
<tr>
<td>Sex</td>
<td>M ...... F...... Other......</td>
</tr>
<tr>
<td>Month and year of birth</td>
<td></td>
</tr>
<tr>
<td>Date of completion of the form</td>
<td></td>
</tr>
</tbody>
</table>
| Household income\(^1\)      | <20000 $....  
20000-40000 $....  
40000-80000 $ ....  
>80000 $ .... |

### Dental Assessment

<table>
<thead>
<tr>
<th>Data</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Missing teeth</td>
<td></td>
</tr>
<tr>
<td>Number of missing teeth in total</td>
<td></td>
</tr>
</tbody>
</table>
| Site of missing teeth       | Maxilla: Anterior....... Posterior.......  
Mandible: Anterior.......Mandible Posterior....... |
| Erupted permanent teeth     |                                  |
| Primary teeth present       |                                  |
| Basic dental examination data: | Mixed dentition......  
Permanent dentition.......  
Overjet ........mm  
Over bite ........% |

For further information regarding how these are categorized please refer to:
Appendix 4: Parent Research Consent Form (The Hospital for Sick Children)

SickKids
THE HOSPITAL FOR
SICK CHILDREN

Research Ethics Board
Parent Research Consent Form (on Behalf of Child)

Title of Research Project:
Oral health related quality of life in children with Oligodontia.

Investigator(s):  
Principal Investigator:  
Dr. Sunjay Suri

Co-Investigators:  
Dr. Leila Raziee

Dr. Peter Judd

Dr. Robert P. Carmichael

Clinical Research Project Coordinator:
**Purpose of the Research:**

We are doing this study because we want to look at the oral health related quality of life in children that have missing teeth (oligodontia). Oligodontia is a condition when a patient has 6 or more missing teeth. We want to get the child’s perspective on any oral symptoms, functional limitations, and emotional and social wellbeing in relation to oligodontia. We also want to get your perspective as the parent of these same things and compare children and parent’s views on the child’s oral health related quality of life.

**Description of the Research:**

All patients with oligodontia who are having an exam to qualify for the Cleft Lip and Palate/Craniofacial Dental Program (CLP/CDP) at the Hospital for Sick Children or Holland Bloorview Kids Rehabilitation Hospital are being invited to participate in this study. If you and your child agree to participate, you and your child will be given two separate questionnaires to fill out. By signing this consent form, you are agreeing to have your child participate in the study. The questionnaires assess the oral health related quality of life in children with oligodontia. They are called Parental-Caregiver Perceptions Questionnaire (P-CPQ) for you to fill out and the Child Perceptions Questionnaire (CPQ) for your child to fill out. You and your child will be asked not to discuss the questions with each other. Your child will also have a dental exam on the same day. This exam will record: missing teeth, number of missing teeth, site of missing, present permanent and primary teeth, and other findings. This exam will only take 5 minutes or less. The missing teeth will be confirmed by X-rays that are available as part of your child’s records in the hospital. Your child will not be exposed to additional X-rays for the purpose of the study. There will be no additional hospital visits as part of this study. The total time for participation in the study will be about 15-20 minutes.

**Potential Harms:**

We know of no harm that taking part in this study could cause you or your child.

**Potential Discomforts or Inconvenience:**

For the purpose of this study, your child will need to spend approximately an extra 15 minutes to complete the questionnaire while you and your child are in the waiting area. The dental exam on your child will take approximately 5 minutes in another clinic room.

**Potential Benefits:**

To individual subjects:
Your child will not benefit directly from participating in this study.
To society:
We will gain a better understanding of the oral health related quality of life of patients with missing teeth. This will help us come up with better management and treatments for these patients.

Confidentiality:
We respect your child’s privacy. No information about your child will be given to anyone or be published without your permission, unless required by law. For example, the law could make us give information about you if a child has been abused, if your child has an illness that could spread to others, if you or someone else talks about suicide (killing themselves), or if the court orders us to give them the study papers.

Sick Kids Clinical Research Monitors, employees of the funder or sponsor, or the regulator of the study may see your child’s health record to check on the study. By signing this consent form, you agree to let these people look at your child’s records. We will put a copy of this research consent form in your patient health record and give you a copy as well.

The data produced from this study will be stored in a secure, locked location. Only members of the research team (and maybe those individuals described above) will have access to the data.

This could include external research team members. Following completion of the research study the data will be kept as long as required then destroyed as required by Sick Kids policy. Published study results will not reveal your child’s identity.

Reimbursement:
Upon completion of the study questionnaires and the clinical examination, two oral hygiene product kits, which include a tooth brush, toothpaste and floss will be provided as compensation for your child’s participation in the study.

Participation:
It is your choice to have your child participate part in this study. Your child can stop at any time. The care your child gets at Sick Kids will not be affected in any way by whether or not your child takes part in this study.

Sponsorship:
The sponsor/funder of this research is the Hospital for Sick Children.
Conflict of Interest:

Dr. Suri and the other research team members have no conflict of interest to declare.

Consent:

By signing this form, I agree that:

You have explained this study to me. You have answered all my questions.

1) You have explained the possible harms and benefits (if any) of this study.

2) I know what I could do instead of having my child take part in this study. I understand that I have the right to refuse to let my child take part in the study. I also have the right to take my child out of the study at any time. My decision about my child taking part in the study will not affect my child’s health care at Sick Kids.

3) I am free now, and in the future, to ask questions about the study.

4) I have been told that my child’s medical records will be kept private except as described to me.

5) I understand that no information about my child will be given to anyone or be published without first asking my permission.

6) I agree, or consent, that my child ________________________ may take part in this study.

Printed Name of Parent/Legal Guardian: ____________________________
Parent/Legal Guardian’s signature & date: ____________________________

Printed Name of person who explained consent: _______________________
Signature of Person who explained consent: ___________________________

Printed Witness’ name: ___________________________________________
Witness’ signature & date: _________________________________________
(if the parent/legal guardian does not read English)

If you have any questions about this study, please call Dr. Sunjay Suri at 416-979-4900 Ext 4397.

If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-813-5718.
Appendix 5: Child Research Consent Form (The Hospital for Sick Children)

Child Research Consent Form

Title of Research Project:

Oral health-related quality of life in children with oligodontia.

Investigator(s):

Principal Investigator:

Dr. Sunjay Suri

Co-Investigators:

Dr. Leila Raziee

Dr. Peter Judd

Dr. Robert P. Carmichael

Clinical Research Project Coordinator:
Purpose of the Research:

We are doing this study because we want to look at the oral health related quality of life in children that have missing teeth (oligodontia). Oligodontia is a condition when a patient has 6 or more missing teeth. We want to get the child’s perspective on any oral symptoms, functional limitations, and emotional and social wellbeing in relation to oligodontia. We also want to get the parent’s perspective of these same things and compare children and parent’s views on the child’s oral health related quality of life.

Description of the Research:

All patients with oligodontia who are having an exam to qualify for the Cleft Lip and Palate/Craniofacial Dental Program (CLP/CDP) at the Hospital for Sick Children or Holland Bloorview Kids Rehabilitation Hospital are being invited to participate in this study. If you and your parent agree to participate, you will be given two separate questionnaires to fill out. These questionnaires assess the oral health related quality of life in children with oligodontia. They are called Parental-Caregiver Perceptions Questionnaire (P-CPQ) for your parent/legal guardian to fill out and the Child Perceptions Questionnaire (CPQ) for you to fill out. You and your parent will be asked not to discuss the questions with each other. You will also have a dental exam on the same day. This exam will record: missing teeth, number of missing teeth, site of missing, present permanent and primary teeth, and other findings. This exam will only take 5 minutes or less. The missing teeth will be confirmed by X-rays that are available as part of your records in the hospital. You will not be exposed to additional X-rays for the purpose of the study. There will be no additional hospital visits as part of this study. The total time for participation in the study will be about 15-20 minutes.

Potential Harms:

We know of no harm that taking part in this study could cause you or your parents.

Potential Discomforts or Inconvenience:

For the purpose of this study, you and your parent will need to spend approximately an extra 10-15 minutes to complete the questionnaires while you are in the waiting area. The dental exam will take approximately 5 minutes in another clinic room.

Potential Benefits:

To individual subjects:
You will not benefit directly from participating in this study.

To society:
We will gain a better understanding of the oral health related quality of life of patients with missing teeth. This will help us come up with better management and treatments for these patients.
Confidentiality:

We respect your privacy. No information about you or your parents will be given to anyone or be published without your permission, unless required by law. For example, the law could make us give information about you if a child has been abused, if you have an illness that could spread to others, if you or someone else talks about suicide (killing themselves), or if the court orders us to give them the study papers.

Sick Kids Clinical Research Monitors, employees of the sponsor, or the regulator of the study may see your health record to check on the study. By signing this consent form, you agree to let these people look at your records. We will put a copy of this research consent form in your patient health record and give you a copy as well.

The data produced from this study will be stored in a secure, locked location. Only members of the research team (and maybe those individuals described above) will have access to the data. This could include external research team members. Following completion of the research study the data will be kept as long as required then destroyed as required by Sick Kids policy. Published study results will not reveal your identity.

Reimbursement:

Upon completion of the study questionnaires and the clinical examination, two oral hygiene product kits, which include a tooth brush, a tooth paste and dental floss will be provided as compensation for your and your parent’s participation in the study.

Participation:

It is your choice to take part in this study. You can stop at any time. The care you get at Sick Kids will not be affected in any way by whether or not you take part in this study.

Sponsorship:

The sponsor/funder of this research is the Hospital for Sick Children.

Conflict of Interest:

Dr. Suri and the other research team members have no conflict of interest to declare.

Consent:

For participants who can consent for themselves:
By signing this form, I agree that:
1) You have explained this study to me. You have answered all my questions.
2) You have explained the possible harms and benefits (if any) of this study.
3) I know what I could do instead of taking part in this study. I understand that I have the right not to take part in the study and the right to stop at any time. My decision about taking part in the study will not affect my health care at Sick Kids.
4) I am free now, and in the future, to ask questions about the study.
5) I have been told that my medical records will be kept private except as described to me.
6) I understand that no information about who I am will be given to anyone or be published without first asking my permission.
7) I agree, or consent, to take part in this study.

Printed Name of Subject & Age                        Subject’s signature & date

Printed Name of person who explained consent       Signature of Person who explained consent & date

Printed Witness’ name (if the subject/legal guardian does not read English)  Witness’ signature & date

If you have any questions about this study, please call Dr. Sunjay Suri at 416-979-4900 Ext 4397.

If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-813-5718.
For parent/legal guardians or substitute decision makers consenting for their children, the consent form must be included with the following final section:

**Consent:**
By signing this form, I agree that:
1. You have explained this study to me. You have answered all my questions.
2. You have explained the possible harms and benefits (if any) of this study.
3. I know what I could do instead of having my child take part in this study. I understand that I have the right to refuse to let my child take part in the study. I also have the right to take my child out of the study at any time. My decision about my child taking part in the study will not affect my child’s health care at Sick Kids.
4. I am free now, and in the future, to ask questions about the study.
5. I have been told that my child’s medical records will be kept private except as described to me.
6. I understand that no information about my child will be given to anyone or be published without first asking my permission.
7. I agree, or consent, that my child ______________________ may take part in this study.

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Printed Name of Parent/Legal Guardian ___________________________ Parent/Legal Guardian’s signature & date

Printed Name of person who explained consent & date ___________________________ Signature of Person who explained consent

Printed Witness’ name ___________________________ Witness’ signature & date
(if the parent/legal guardian does not read English)

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If you have any questions about this study, please call Dr. Sunjay Suri at 416-979-4900 Ext 4397.

If you have questions about your rights as a subject in a study or injuries during a study, please call the Research Ethics Manager at 416-813-5718.
Appendix 6: Research Assent Form (The Hospital for Sick Children)

Research Assent Form

**Title of the Research Project:**

Oral health-related quality of life in children with Oligodontia.

**Investigator(s):**

Principal Investigator:
Dr. Sunjay Suri

Co-Investigators:

Dr. Peter Judd

Dr. Robert P. Carmichael

**Why Are We Doing This Study?**

We are doing this study because we want to know how you and your parents feel about you having some missing teeth.

**What Will Happen During The Study?**

You are your parents will be asked to fill out a form about your missing teeth. We will also look into your mouth to count your teeth.

**Are there good things and bad things about the study?**
Nothing bad will happen during the study. The good thing is we will know about your teeth and how you feel.

**Who will know that I did the study?**
No one else will know that you did this study. If we feel your health may be in danger, we may have to report your results to your doctor.

**Can I decide if I want to be in the study?**
Nobody will be angry or upset if you do not want to be in the study. We are talking to your parent/legal guardians about the study and you should talk to them about it too.

**Assent:**
"I was present when ______________ read this form and said that he or she agreed, or assented, to take part in this study.”

______________________________
*Printed Name of person who obtained assent*

______________________________
*Signature & Date*
Appendix 7: Child/Parent Consent Form (Holland Bloorview Kids Rehabilitation Hospital)

Study Title: Oral health-related quality of life in children with Oligodontia.

Principal Investigators:
Dr. Robert P. Carmichael
Dr. Sunjay Suri
Co-Investigators:
Dr. Leila Raziee
Dr. Peter Judd

Sponsor/Funder(s): Holland Bloorview Kids Rehabilitation Hospital, The Hospital for Sick Children

Dear Participant/Caregiver,

My name is Leila Raziee. I am part of a research team at the Bloorview Research Institute at Holland Bloorview and we want to look at the oral health related quality of life in children that have missing teeth (oligodontia). Before agreeing to take part in this study, it is important that you understand how you/your child will be involved. This consent form gives information to help you/your child make an informed choice.
What is the study about?

We are doing this study because we want to look at what the quality of life is like in children that have missing teeth (oligodontia). We want to get the child’s perspective on any issues about their mouth as well as their feelings in relation to having oligodontia. We also want to get the parent’s perspective of the same things and compare children and parent’s views.

We are planning to include 35 pairs of children with missing teeth (oligodontia) and their parents. About 20 patients and their parent will be selected from Holland Bloorview and 15 patients and their parents from The Hospital for Sick Children (SickKids). We want to invite you/your child to be one of the families for this study.

How will my child and I be involved in this study?

All patients with oligodontia who are having an exam for the Cleft Lip and Palate/Craniofacial Dental Program (CLP/CDP) at Holland Bloorview Kids Rehabilitation Hospital or the Hospital for Sick Children are being invited to participate in this study. If you and your child agree to participate, you each will be given two separate questionnaires to fill out. These questionnaires assess the oral health related quality of life in children with oligodontia. They are called Parental-Caregiver Perceptions Questionnaire (P-CPQ) for the parent to fill out and the Child Perceptions Questionnaire (CPQ11-14) the child to fill out. While completing the questionnaires you/your child will be asked not to talk about the questions with each other. The parent will also be asked to fill out a short questionnaire about the family demographics. The child will also have a brief dental exam right after. This exam will record some things already recorded during their appointment as well as some extra information on their teeth. This exam will only take 5 minutes or less. The missing teeth will be confirmed by X-rays that are available as part of the child’s record in the hospital. The child will not be exposed to any more X-rays for the purpose of the study. There will be no extra hospital visits as part of this study. The total time for participation in the study will be about 15-20 minutes.

Will anyone know what I say?

We will take the filled out questionnaires from the parent and the child. We respect your/your child’s privacy. No information about you (parent or the child) will be given to anyone or be published without your permission, unless required by law.

Holland Bloorview Clinical Research Monitors or the regulator of the study may see your/your child’s health record to check on the study. By signing this consent form, you agree to let these people look at your/your child’s records.

The data we get from this study will be stored in a secure, locked location. Members of the research team will have access to the data. Holland Bloorview representatives and the Holland Bloorview Research Ethics Board (REB) will be allowed to see the research records for making sure research procedures and data collection are following the hospital policies. The records will be confidential, to the extent permitted by the applicable laws and regulations as per REB SOPs.
Research data will be kept for 7 years after the study is finished and then securely destroyed/deleted. Published study results will not reveal your identity.

**Do I have to do this?**

It is your choice to take part in this study. You can stop at any time. The care you/your child gets at Holland Bloorview will not be affected in any way by whether or not you take part in this study.

**What are the risks and benefits?**

We know of no harm that taking part in this study could cause you or your child. However, it may cause emotional distress while answering some of the questions. These are questions (12-20) on the child CPQ which address feelings about having oligodontia and questions about spare time activities and being with other people. In the parents CPQ, it is questions 17-49. If this does happen, we will give you support by directing you to social services and counselling in the hospital.

For the purpose of this study, the parent and the child will need to spend approximately an extra 10-15 minutes to complete the questionnaires. The dental exam will take approximately 5 minutes.

You will not benefit directly from participating in this study. We will gain a better understanding of the oral health related quality of life of patients with missing teeth. This will help us come up with better treatments for these patients. You can receive a copy of the study results at the end of the study.

**Do the investigators have any conflicts of interest?**

I, Leila Raziee and the other research team members have no conflicts of interest to declare related to this study.

**Reimbursement:**

Once you have completed the questionnaire and dental exam, we will give both the parent and child a tooth brush, a tooth paste and dental floss to show our appreciation for your participation in the study.

**What if I have questions?**

Please call Dr. Robert Carmichael to explain anything you don’t understand before signing the consent form. His phone number is 416-425-6220 ext 3872. If you leave a message, he will return your call within 48 hours.

You will get a letter at the end of the study to learn about the study findings.

If you have any questions about your rights as a research participant, please contact the Holland Bloorview Research Ethics Board at 416-425-6220 ext. 3507.
INFORMED CONSENT FORM
HOLLAND BLOORVIEW KIDS REHABILITATION HOSPITAL

Re: Oral health-related quality of life in children with oligodontia.

Please complete this form below and return it to the researcher. You will receive a signed copy of this form.

By signing this form, I confirm that:
- Leila Raziee explained this study to me and answered all of my questions.
- I read the attached Informed Consent Form dated (June 14 2017) and understand what this study is about.
- I understand the known risks and benefits of participating in this research study.
- I understand that I may drop out of the study at any time. My decision about taking part in the study will not affect the services the child gets at Holland Bloorview.
- I am free now, or in the future to ask questions about the study.
- I know that study records related to the child will be kept confidential except as described in this form.
- I understand that information that identifies the child or family will not be shared with anyone without first asking my permission.
- I agree to participate in this study and allow/support the child’s decision to participate.

Participant’s Name ___________________________ Signature ___________________________ Date __________
(please print)

I have explained this study to the above participant and have answered all their questions.

Name of Person Obtaining Consent ___________________________ Signature ___________________________ Date __________
Appendix 8: Assent Form (Holland Bloorview Kids Rehabilitation Hospital)

Title: Oral health-related quality of life in children with Oligodontia.

Why did you want to talk with me?
We are doing this study because we want to know how you and your parents feel about you having some missing teeth.

What do I need to do?
You are your parents will be asked to fill out a form about your missing teeth. We will also look into your mouth to count your teeth.

What are the good and bad things about doing this?
Nothing bad will happen during the study. The good thing is we will know about your teeth and how you feel.

Will anyone know that I did this study?
No one else will know that you did this study. If we feel your health may be in danger, we may have to report your results to your doctor.

Do I have to do this?
Nobody will be angry or upset if you do not want to be in the study. We are talking to your parent/legal guardians about the study and you should talk to them about it too.

What if I am not sure?
Your parents know about this study. Ask them questions if you don't understand what this is about. You can also talk to me about the study before you decide whether or not you want to be involved.

Thanks for thinking about being a part of this study.
Oral health-related quality of life in children with Oligodontia.

I discussed this form with ____________________________ and s/he agreed to participate.

Name of Person Who Obtained Assent

Signature

Date
Appendix 9: REB Approval (The Hospital for Sick Children)

Research Ethics Board (REB) Study Approval Letter

2017-06-09

Sunjay Suri Dentistry

REB number: 1000055472
Study Title: Oral health-related quality of life in children with oligodontia.

Date of Approval: 2017-06-09
Expiry Date: 2018-06-09

Thank you for the application submitted on 2017-05-01. The above referenced study was reviewed through a delegated process (not by Full Board review). Any concerns arising from this review have been documented and resolved.

The REB voted to approve this study, and your participation as Principal Investigator, as it is found to comply with relevant research ethics guidelines, as well as the Ontario Personal Health Information Protection Act (PHIPA), 2004.

The Hospital for Sick Children Research Ethics Board hereby issues approval for the above named study. This approval is effective from 2017-06-09 to 2018-06-09. Continuation beyond that date will require further review of REB approval.

The following documents have been reviewed and are approved:

2. Assent: Version 2 – June 6, 2017 [Assent Form June 6 2017_Clean.docx (1.0)]
3. Participant Consent: Version 2 – June 6, 2017 [Child Consent Form June 6 2017_Clean.docx (1.0)]
5. Parent Consent as Participants: Version 2 – June 6, 2017 [Parent Consent Form May 15 2017_parentquestionnaire_Clean.docx (1.0)]

During the course of this investigation, any significant deviations from the approved protocol and/or unanticipated developments or significant adverse events should immediately be brought to the attention of the REB.

The SickKids REB operates in compliance with the Tri-Council Policy Statement; ICH Guideline for Good Clinical Practice E6(R1); Ontario Personal Health Information Protection Act (2004); Part C Division 5 of the Food and Drug Regulations, Part 4 of the Natural Health Products Regulations and the Medical Devices Regulations of Health Canada. The approval and the views of the REB have been documented in writing. The REB has reviewed and approved the clinical trial protocol and informed consent form for the trial. All investigational drug trials at SickKids are conducted by qualified investigators.

Furthermore, members of the Research Ethics Board who are named as Investigators in research studies do not participate in discussions related to, nor vote on such studies when they are presented to the REB.

REB # 1000055472
Research Ethics Board (REB)
Renewal Approval Letter

2018-06-01

Dr. Sunjay Suri
Dentistry

REB number: 1000055472
Study title: Oral health-related quality of life in children with oligodontia.
Date of initial study approval: 2017-06-09
Study expiry date: 2019-06-09

Thank you for your renewal application requesting a renewal and approval of the above named study.
This letter will serve as an extension of the SickKids Research Ethics Board (REB) approval for the study. This renewal was approved by the REB via delegated review (not by Full Board review). This approval is effective from 2018-06-09 to 2019-06-09. Continuation beyond that date will require further review of REB approval.

During the course of this investigation, any significant deviations from the approved protocol and/or unanticipated developments or significant adverse events should immediately be brought to the attention of the REB.

The SickKids REB operates in compliance with the Tri-Council Policy Statement; ICH Guideline for Good Clinical Practice E6(R1); Ontario Personal Health Information Protection Act (2004); Part C Division 5 of the Food and Drug Regulations; Part 4 of the Natural Health Products Regulations and the Medical Devices Regulations of Health Canada. The approval and the views of the REB have been documented in writing. The REB has reviewed and approved the clinical trial protocol and informed consent form for the trial. All investigational drug trials at SickKids are conducted by qualified investigators.

Furthermore, members of the Research Ethics Board who are named as Investigators in research studies do not participate in discussions related to, nor vote on such studies when they are presented to the REB.
Appendix 10: REB Approval (Holland Bloorview Kids Rehabilitation Hospital)

Dear Dr. Carmichael,

The Holland Bloorview Research Ethics Board (REB) has reviewed the above named study. This was a delegated review. The board is granting ethics approval for a period of one year. The approval of this study includes the following documents:

- Protocol (version 5 dated August 23, 2017)
- TAHSN Form received August 17, 2017
- Informed Consent Form – Participant/Caregiver (version 3 dated August 17, 2017)
- Assent Form (version 1 dated March 14, 2017)
- Capacity Assessment Tool (version 1 dated August 17, 2017)
- Participate in Research Recruitment Fyer (version dated June 14, 2017)
- Recruitment Script (version received May 19, 2017)
- Oligodontia Data Collection Form (version 3 dated August 17, 2017)
- Child Oral Health Questionnaire (version 1 dated June 6, 2017)
- Parent-Caregiver Perceptions Questionnaire (version 1 dated June 6, 2017)
- Thank you Letter (version 1 dated June 14, 2017)
- Code Breaking File (version 1 dated June 14, 2017)

This study must be conducted in accordance with the description in the application and any supplementary documents for which ethics approval has been granted. The REB needs to be notified of any unanticipated or unintentional divergence or departures from the protocol through a "Protocol Deviation Form". Any intentional changes to the protocol need be submitted through an "Amendment Form" to the REB for approval before the changes are implemented, except where necessary to eliminate immediate hazards to the participants.

Any adverse events that occur as a result of your study must be reported to the REB by submitting an "Adverse Event/Unanticipated Problem Form". If the study is expected to continue beyond the new expiry date, you must request another renewal, at least thirty days prior to the expiry date, by submitting an "Annual Renewal Form". When the study is completed or terminated, you need to submit a "Study Closure Form" to the REB.

Best wishes for the successful completion of your project.