An evaluation of the caries experience among individuals with Down syndrome

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ABSTRACT

Objective: To evaluate the caries experience of people with Down syndrome (DS) compared to people without DS. Methods: A cross-sectional study was conducted. The caries experience was assessed by a crude DMFT score and an adjusted DFT score expressed as a percentage of caries-affected teeth to compensate for hypodontia in the DS group. Multiple logistic and linear regressions were used to compare the outcomes of both groups, controlling for all predictors. Results: Of the 44 DS and 84 non-DS subjects, the adjusted DFT score showed non-significant differences between them (p=0.18). The crude DMFT score displayed borderline significance (p=0.06), while people without DS had a higher likelihood of a caries experience compared to the DS group (OR, 4.6; 95% CI, 1.5-14.0). Conclusions: Independent of other predictors, the caries experience was greater among people without DS, without adjusting for the number of teeth. However when this factor is considered, the caries experience is the same among both groups.

RÉSUMÉ

Objectif: Évaluer la prévalence de la carie dentaire des trisomiques (T21) comparativement à celle des non trisomiques. Méthodologie: Une étude transversale a été conduite. La prévalence de la carie dentaire a été mesurée par l'indice CAOD et par un indice COD ajusté exprimé en pourcentage de dents affectés par la carie, pour compenser pour l'hypodontie parmi les T21. Des régressions logistique et linéaire multiples ont été effectuées. Résultats: Pour les 44 sujets T21 et les 84 sujets non T21, aucune différence significative a été démontrée par l'indice COD ajusté (p=0.18). L'indice CAOD brut était presque significatif (p=0.06) où la chance de développer la carie dentaire était plus élevée parmi les non T21 que pour les T21 (OR, 4.6; 95% CI, 1.5-14.0). Conclusion: Indépendamment des autres prédicteurs, la prévalence de la carie dentaire était plus élevée chez les non T21, sans ajustement du nombre de dents. Par contre en ajustant ce facteur, le score est le même entre les deux groupes.

1. INTRODUCTION

This past century has witnessed remarkable advances in the understanding of individuals with Down syndrome (DS). At the turn of the twentieth century, a person with DS was expected to live to age nine, while living past fifty is not uncommon today. These improvements reflect consequences of changes in lifestyle, renewed opportunities, and vigorous health research. Nevertheless, minimal emphasis has been directed towards this group's oral and dental health, and questions continue to linger at the dawn of the twenty-first century.

Recent literature concerning the dental health of individuals with DS is scarce. In addition, information pertaining to the caries experience of this population is further confused with conflicting results from past studies. In accordance with supporting literature, it is widely accepted that individuals with DS exhibit a higher prevalence of dental disease, such as periodontitis and gingivitis, and poorer oral hygiene compared to the general population.[1] Yet paradoxically, there has been less agreement on the prevalence of dental decay among these individuals.

The results concerning the caries experience among people with DS are mixed. Numerous studies have reported that individuals with DS exhibit fewer dental caries, compared to other challenged groups and the general population.[2-17] However other studies have demonstrated differing results. A few findings have shown that there is no difference in the caries experience between people with and without DS,[18-22] while others have found a higher prevalence of dental decay among individuals of the DS group.[23-25]

While keeping these ambiguous results in mind, two additional factors drive the need for continued research on the caries experience among people with DS. The first factor embodies methodological limitations of past studies, and the second illustrates sociobiological changes that have impacted this particular population over time.

With respect to the first issue, many studies exhibit biases and inaccuracies in the study design, compromising their accuracy and generalizability to the DS population. A few review articles have observed that many older studies have been poorly controlled, neglecting fundamental physiological differences between people with DS and the general population.[4, 17, 26] For example it is widely recognized that congenitally missing teeth ranging from a few teeth (hypodontia) to many teeth (oligodontia) are highly prevalent among people with DS compared to the non-DS population. Yet despite this wide acknowledgement, most studies, ironically, rarely take into account the differences in the number of teeth when the two groups are compared. There has been scepticism and discussion that congenitally missing teeth and delayed eruption may contribute to the lower caries experience among people with DS, but only a few authors have applied this issue in their own studies.

To date, only four out of eighteen epidemiological studies have explicitly taken the differences in the number of teeth between people with and without DS into consideration when calculating the caries experience.[12, 19, 22, 27] Therefore, because the decayed, missing, filled teeth (DMFT) and decayed, missing, filled surfaces (DMFS) scores are dependent on the number of teeth, ignoring such differences jeopardizes the accuracy of comparing the caries experience between both groups.

In addition, many studies have either failed to control or inappropriately categorize individuals with respect to their developmental disabilities [26] and living arrangements.[4] Both factors vary among the subject population and exhibit different dental care practices, significantly affecting the caries experience. Finally, many studies have exhibited low sample sizes, compromising the statistical power and the reliability of the results.[5, 11, 13, 22, 28, 29]

The final rationale for further inquiry into the caries experience of people with DS involves changes in the sociobiological context of this population. First, the incidence of DS is expected to remain stable in Canada and the US.[30, 31] Second, the life expectancy of individuals with DS has increased by nearly 1.7 times since the early 1980s.[32] And lastly, most studies that pertain to the oral health of individuals with DS are dated, and the social development of this cohort has dramatically changed in the last decade. A majority of the studies were published between the 1960s until the early 1980s, reflecting a different social circumstance amongst this population today.

Changes in social policy to abolish systematic discrimination against the physically and cognitively challenged has recently occurred in the United States,[33] while a landmark resolution was made within the United Nations body.[34] With these laws and resolutions in place, changes in socio-economic policies towards the normalization and integration of challenged individuals into society have provoked changes in mainstream society's values. The lifting of barriers and stigmas against this population has stimulated notable improvements in education and lifestyles. Many may agree that such alterations in social policies pose a significant influence on a person's health and well-being. Since oral health is an integral part of total overall health, it may be reasonable to expect that alterations in social policies among this group may have an important influence on a person's oral health.

Therefore because of the aforementioned study limitations and the sociobiological changes that have affected the DS population, the importance of determining the caries experience of this group must not be underestimated. This significance is also further exemplified with little caries research among this group from a Canadian context. In light of these factors, this thesis will address these issues, while attempting to control for as many factors that are significant and applicable to people with DS today. More specifically, the primary objective of this study is to evaluate the caries experience of individuals with DS compared to age- and dentist-matched control subjects without DS.

In accordance with the specifications approved by the Graduate and Postdoctoral studies at McGill University, a review of the literature will be presented in chapter two, the objective and hypotheses in chapter three, and a detailed description of the methodology in chapter four. In addition the results, discussion and conclusions will be illustrated in chapters five, six, and seven, respectively.

2. LITERATURE REVIEW

2.1. Epidemiology

Individuals with DS have intrigued many in the last 150 years. Not only did they capture the attention of John Langdon Down who was the first to describe and document their phenotype in 1866, but this population is still in the light of much health and social research today.

To date, DS remains the most common cause of genetic intellectual disability among humans. More specifically this autosomal chromosomal abnormality represents approximately 20% of the intellectually disabled population. Current information on the prevalence of DS in Canada is unavailable. However in the United States, this disability affects over 350,000 individuals.[33]

In 1999, DS constituted 12.5 per 10,000 live births and 14.2 per 10,000 total births in Canada.[31] The incidence of DS varies from country to country, depending on the cultural and social context of the country, and the legality of terminations of pregnancy. For example in the same year, 4.9 per 10,000 live births were recorded in Central East France where elective terminations are common, and 18.0 per 10,000 live births were recorded in the United Arab Emirates where this procedure is illegal.[31]

Yet despite recent and notable differences on the incidence of DS among various countries, long-term predictions in Canada remain stable. With the increase in average maternal age, earlier sources have estimated that the birth prevalence of DS would remain constant in developed nations because of the advances in prenatal diagnosis that enable early detection of this disability.[30, 35] Such trends are reflected in Canada, where the birth prevalence rate has remained stable at approximately 13.2 per 10,000 total births between 1989 and 1999.[31] Provincial and territorial variation exists, with the exception of Alberta showing an increased trend from 0.89 per thousand births in the early 1980s to 1.11 per thousand births in the early 1990s.[36] In this province, this trend is expected to increase over time despite the rise of prenatal genetic testing.[37]

As the birth prevalence of DS is expected to remain generally constant over time in Canada, it is anticipated that there will be an increase in the overall prevalence rate. It has been observed in the last two decades that there has been a substantial increase in the life expectancy of individuals with DS. Of the 17,897 subjects in a recent study by Yang *et al.*,[32] the median age at death increased from 25 years in 1983 to 49 years in 1997. Although the median life expectancy has increased 1.7-fold, it has been shown that there are remarkable disparities between different racial groups in the United States. There is currently no published data available on the life expectancy of people with DS in Canada.

2.2. Aetiology

In 1959, scientists published the genetic basis underlying the unique characteristics of individuals with DS.[38] These individuals were found to possess a normal set of 46 chromosomes, in addition to an extra copy of chromosome 21. As a result of this duplication, it was observed that this population has a total of 47 chromosomes. Trisomy 21 is an alternate name for DS, and this particular term derives from the fact that there are three copies of chromosome 21, and is the site where gene expression gives rise to this group's distinctive phenotype.[33] Above all, it is found that the over-expression of the genes on band 21q22.1 poses the most significant influence on the health and physical traits of this cohort.[39]

There are three main ways by which chromosome 21 can be duplicated.[40] In 94% of all cases of DS, an additional chromosome is formed due to an error in cell division during the first stage of meiosis. Namely, this process of non-disjunction (also called, *Standard Trisomy 21*) occurs when one pair of chromosome 21 fails to separate, giving rise to three copies of this chromosome within all the cells of the offspring. Alternatively somatic mosaicism (also called, *Partial Trisomy 21*) accounts for approximately 2% of all cases, whereby the body's cells contain a mixture of either 47 chromosomes and either a normal set of 46 chromosomes. Both non-disjunction and mosaicism are genetic means that are not inherited from either parent. On the other hand, the last method involving an unbalanced translocation of chromosome 21 onto another different chromosome occurs in about 4% of all cases. Approximately one-third of these cases are inheritable.

The causes of having a child with DS are multifactorial in origin.[31] Environmental factors such as medical irradiation, pesticides, and alcohol, in addition to genetic anomalies play a combined causal role in this disability. Although there are many interacting factors leading to the development of Trisomy 21, it is interesting to note that there is new evidence showing that nutritional status may contribute to this disorder.[41] Folic acid intake is seen to positively correlate with a decreased risk of having a child with DS. This nutrient is found to regulate folate pathway genes that prevent or decrease the likelihood of meiotic non-disjunction.

Nevertheless among all known predisposing factors, advanced maternal age is by far the strongest.[42] The risk of having a baby with DS increases with the mother's age, regardless of racial, social, economic, cultural or religious background. For example, a mother who is 23 years old has a one in 1447 chance of having a baby with DS, while the risk is one in 65 by age 42.[43] This exponential increase in risk is also reflected on the prevalence of DS in Alberta between 1990 and 1998.[31] For mothers between 25 and 29 years, the prevalence of DS is 7.2 per 10,000 total births, and it increases to 28.3 per 10,000 total births for mothers between the ages of 35 and 39. Although the risk of having a child with DS increases significantly with age, approximately 80% of all cases are born to mothers under 35 years. Such phenomenon occurs since most childbearing rests below this age.

2.3. Advances in public policy advocacy

Aside from improvements in health research among people with DS, significant advances in public policy concerning this population have also occurred. Such changes have been shown to influence oral health.[3]

The increasing trend towards the normalization and integration of people with DS and other developmental challenges into the general population have resulted from two important civil rights legislations.[33] In 1975, the *Individuals with Disabilities Education Act* (IDEA) in the United States was passed and then amended in 1990. This law dictated the "right to an individualized, free, and appropriate public education" to all people with disabilities.[33] The second important legislation affecting this cohort was a 1992 United Nations resolution.[34] This declaration formally removed social and physical barriers encountered by the cognitively and physically challenged.

As a result of these formal changes in social policy, increasingly more challenged individuals have had the ability to access the domains of everyday life, and are living independently in the community, or at home with their family.[33, 34] However as the level of independence increases, access to proper dental treatment has unfortunately decreased. Recent studies have shown that most people with DS have difficulty accessing a dentist due to social stigmas, financial barriers, difficulties in physical access, and limited number of dental professionals willing to care for people with special needs.[44-47]

2.4. Anatomical and systemic considerations

The uniqueness of DS is demonstrated by variability in the physiological and anatomical aspects of this population. Not all individuals with DS share the same problems or conditions. However, this section will provide a brief overview of the most common anatomical difficulties and comorbidities that are observed in this population.

2.4.1. Anatomical considerations: Orofacial characteristics

The over-expression of chromosome 21 brings about many distinguishing phenotypes. From an anatomical perspective, people with DS are characterized with unique craniofacial features. The head is generally smaller in size (microcephaly), while the skull is often short and broad (brachycephaly) with a flattened occiput.[1] Compared to the mandible, the maxilla is usually underdeveloped, which results in a palate with reduced length, height and depth.[48, 49] As such, the palatine arch becomes high and oval-shaped.

The underdeveloped maxilla also gives rise to poor dental occlusion. Pseudo-class III malocclusion, characterized by an anterior crossbite with a forward mandibular displacement, is most observed in this population.[50] Such development is thought to be due to the abnormal tongue position. However, other abnormal jaw relationships are also present among people with DS. Approximately 69% experience a mandibular overjet, an anterior open bite is observed in about 54% of all cases, 97% possess posterior cross bites, 65% with a mesial molar occlusion, and most develop an anterior cross bite.[51]

In addition, common facial characteristics consist of a flattened nasal arch accompanied by a wide nasal root and high nasal tip.[1] With respect to the eyes, there is generally an excess width between them (hypertelorism), with the presence of up-slanting and short eyelids. Epicanthal folds are also present, in which there is a skin fold of the upper eyelid covering the inner corner of the eye.[10] Such folds often regress with age.

It is also important to note the unique dental and oral features of this population. Clinically, people with DS are present with a small oral cavity (pseudomacroglossitis) and protruding, macroglossic tongue.[48] Together with an abnormal sized tongue and oral cavity, an underdeveloped maxilla, hypotonic lower lip and large tonsillar volume promote the tongue to rest on the lower teeth to allow a clear airway for breathing in a rested state.[1] Such approach to respiration commonly causes the fissuring of the tongue and lips. Xerostomia is also commonly observed as a result of the combination of chronic mouth breathing and the thinning of the oral mucosa.[52]

Regarding dental anomalies, many dental malformations exist amongst this cohort. Approximately 50% of all people with DS have congenitally missing teeth ranging in severity from hypodontia to oligodontia. The upper lateral incisors are in general the most affected.[53, 54] In addition, the morphology and structure of the teeth are unique. Between 35 to 55% of all people with DS have abnormally small teeth (microdontia).[48] Microdontia is clinically seen in all teeth except for the upper first molars and lower incisors. Additionally, due to the small size of most teeth, spacing is also very common.[52] Furthermore the clinical crowns are frequently conical and shorter in shape, leading to a decreased root to crown ratio.[48] Eruption times are often delayed among primary and permanent dentitions, lasting for as long as two to three years,[55] while its eruption sequence is generally abnormal.[1]

2.4.2. Clinical findings: A systemic perspective

Individuals with DS are prone to many medical conditions, in addition to the many physical abnormalities summarized above. Recent studies show that many systemic diseases share common risk factors with oral diseases.[56] Thus such systemic dysfunctions observed among this population may closely relate to the development of oral diseases such as dental caries. Oral diseases, as a result, may further intensify systemic conditions.[1] In light of these potentially interacting risk factors, this section will provide a brief overview of the common medical problems shared among this population.

Approximately 40 to 50% of all people with DS are born to experience a form of congenital heart disease.[57] There are abnormalities of the atrial septum, the atrioventricular valves and the ventricular systems, however defects in the endocardial cushion are the most common.[58] By adulthood, nearly half of all people with DS develop Mitral Valve Prolapse (MVP). This is a very high incidence rate compared to the general population in which 5 to 15% are affected.[48,59] Among these patients with MVP, most undergo antibiotic prophylaxis for bacterial endocarditis as a dental treatment since it has been found that the combination of peridontitis and poor oral hygiene increases the risk of developing this cardiac infection.[59]

Anomalies with the nervous system must also be stressed. Neurological disease is highly prominent among people with DS, where many are affected by a wide degree of mental retardation. Specifically, individuals over 35 years are at high risk of Alzheimer's disease, with the incidence nearly twice that of the general population.[60] The onset of clinical symptoms usually appear between the ages of 40 to 50 years, where 70% exhibit no clinically detectable behavioural changes.[52] In addition, the risk of epilepsy increases with age.[61]

From a musculoskeletal standpoint, reduced muscle tone, or muscle hypotonia, occurs throughout the body in a rested state. Such dysfunction also affects the smooth muscle of the gastrointestinal tract and is a distinguishing feature amongst the DS population.[33] Poor motor control, especially in the lips, tongue and cheeks, compromises oral function such as chewing efficiency, speech, suckling, swallowing, and the natural cleansing of the teeth.[1, 54] As a result, the underdeveloped motor coordination also affects the dental and nutritional aspects of the individual. Normal feeding is at risk due to problems in swallowing and mastication,[62] and with respect to dental care, there is a decreased level of manual dexterity manifesting poor brushing and flossing skills.[54]

Such reduction in muscle tone not only delays gross motor development, but it is also associated with generalized ligamentous laxity. Joints are commonly unstable, affecting areas such as the temporomandibular joint (TMJ) ligaments. Also approximately 20% of all cases are affected by atlanto-axial instability between the C1 and C2 vertebrae.[63] Laxity in this area brings about difficulties with swallowing. The head generally has a tendency to lean back, fostering a blocked oropharynx by the tongue.[1]

Individuals with DS also experience hematopoietic anomalies. First, many possess a compromised immune system. Neutrophil leukocytes are defective and short lived, while cell mediated immunity is impaired and serum immunoglobin patterns are disrupted.[48] As such, the decreased ability of the T-cells to fight disease and a systematic decrease in immunoglobulins together contribute to an increased susceptibility to infections.[33] Common infections include periodontal disease, oral candida, upper respiratory, and middle-ear infections.[54] In addition, there is a high incidence of leukemia among young children with DS. While 1 in 200 are affected, this rate is between 10 to 15 times greater in this group, compared to the general population.[52]

2.4.3. Clinical findings: An oral/dental perspective

2.4.3.1. Bruxism

Bruxism is very common among children and adults with DS. The extent of tooth grinding varies among each individual. This condition may be mild, potentially wearing away some of the secondary and tertiary fissures and grooves among newly erupted teeth. In more acute cases, it may be severe enough to break down the supporting tissues of the teeth.[48] Nevertheless the justifications for bruxism remain inconclusive; the current belief is that it is caused by multiple factors such as severe dental malocclusion, TMJ dysfunction, and an underdeveloped nervous control.[1] However more recently, there has been increasing evidence that bruxism may be due to the modulation of the central nervous system (CNS). Although the specific areas of the CNS that cause bruxism are still debated, one study showed that the regulation is in the central dopaminergic system.[64] Another paper has further refined the results of the latter study, showing that the potential source of this problem is in the limbic system that controls emotions [65]– a biological finding that parallels other studies arguing that tooth grinding is partially due to the chronic stress that many people with DS experience.[54]

2.4.3.2. Periodontal Disease

With respect to dental diseases, it is widely known that people with DS have a higher prevalence of periodontal disease compared to other mentally challenged individuals and the general population.[5, 9, 22, 66] Specifically, this disease affects nearly 94% of adolescents between the ages of 16 and 20.[1]

Clinically, the extent of gum disease resembles that of juvenile periodontitis.[67] In addition marginal gingivitis, advanced periodontitis, gingival recession and pocket formation are frequently observed among people with DS.[68] In general mandibular incisors and maxillary molars are the most affected.[67]

The causes for periodontal disease encompass several factors, such as poor oral hygiene, poor masticatory function, dental imbrication, and an impaired immunological response.[68-70] With respect to the latter factor, a 7-year intervention study by Cichon *et al.* [68] has demonstrated the significant role of the impaired immune system on the development of periodontitis among people with DS. This cognitive impairment and age-matched study has found that the development of periodontal disease is independent of the amount of plaque found on the teeth. The authors have subsequently reported that the progression of this disease is primarily due to their lowered immune host response.

Although the direct causes of periodontitis remain uncertain among individuals with DS, there is firm belief that the periodontal breakdown is rapid and severe. However more recently, its severity and progression was found to be slower than expected among the adult DS population.[66] This result may indicate that the clinical dental status of people with DS may be improving due to increased interventions and research in the clinical and basic sciences.

2.4.3.3. Caries

Although people with DS are certainly more likely to experience periodontal disease than individuals without DS, the caries experience remains unclear. Dental decay is a multifactorial disease.[17] Its development and progression varies according to factors such as, dietary habits, fluoride intake, cooperation levels, age, dental care visits, gender, oral hygiene, drug use, microbiological factors, and genetics. Because its causes are multiple, deciphering the caries experience of people with DS is made more difficult due to their unique physiological and social challenges (see sections 2.4.1 - 2.4.3.).

Thus as indicated in the introduction, consensus on the caries experience of people with DS is discordant and conflicting. A couple of studies have reported a high prevalence of disease.[23, 24] For example in a cross-sectional study by Shyama *et al.*,[23] the caries experience (as measured by a DMFS score) was found to be significantly higher among people with DS, when compared to other developmentally challenged subjects ($p=0.03$ for primary dentition, and $p<0.001$ for permanent dentition). This finding was consistent among the entire study sample, ranging from 3 to 29 years.

However, five studies have found that the caries experience is the same among people with and without DS.[18-22] Among them is a large-scale study by Cutress.[19] This was a historical cohort study involving 416 subjects with DS, 432 cognitively challenged subjects, and 697 control subjects without any disabilities, in which the age range was between 5 and 24 years. The author expressed the caries experience in numerous ways; however one noteworthy approach was the DMFT expressed as a percentage, with respect to the total number of teeth. The author found that there were no differences in the caries experience among all three subject groups, however this differed when the population was subdivided by living arrangement. Individuals who lived in institutions had a significantly lower caries experience than individuals who lived at home with at least a parent or guardian.

In addition, a more recent age- and gender-matched cross-sectional study by Ulseth *et al.* has demonstrated similar results among an adult population between the ages of 21 and 72 years.[22] To compensate for the high number of missing teeth among the DS subjects, the author followed similar approaches to Cutress to standardize the DMFT score – however the number of missing teeth due to decay were not taken into consideration in the calculations. The investigators' devised *DFT/DFST ratio* provided a comparison of the caries experience with other mentally challenged individuals. Specifically this is a ratio that divides the number of decayed and filled teeth, with the total number of teeth present at the time of examination. It was found that approximately 50% of all teeth were affected by caries in the DS group, while 48% were affected by caries in the non-DS group. This difference was statistically non-significant, concluding that the caries experience was similar between both groups.

Yet despite the seemingly persuasive studies, an overwhelming number of papers have reported a lower caries experience when compared to the non-DS population.[2-17, 19] Stabholz *et al.* found that children with DS between the ages of 8 and 13 had significantly lower DMFS scores than other cognitively challenged and non-challenged children of the same age group.[13] The DS group resulted in an average DMFS of 1.2, while non-challenged children had a score of 14.5, and the challenged group with a DMFS of 15.6. (All differences were highly significant at $p<0.01$). The authors have suggested that this difference may be due to many factors distinctive to the DS group, such as delayed tooth eruption, reduced time of exposure to a cariogenic environment, lower bacterial counts of *Streptococcus mutans*, higher salivary pH and bicarbonate levels. In addition, similar results were found in a one-year longitudinal study involving 132 subjects, with 32 being in the DS group.[3] The DS group possessed a mean DMFT of 8.1 while the non-DS group had a mean DMFT of 18.2. The difference in DMFT in these two groups was statistically significant (p=0.002).

Nevertheless, in four out of eighteen epidemiological studies, the unusually high number of congenitally missing teeth and delayed tooth eruption were overtly taken into consideration in the calculation of the caries experience.[12, 19, 22, 27] Most studies have acknowledged and discussed this physiological difference between the DS and non-DS population, representing an important factor in influencing the calculation of the caries experience. Failing to take into consideration each person's total number of teeth leads to incomparable results. Calculations made to explore the caries experience, whether employing the DMFT or DMFS, are all dependent on the total number of teeth in the mouth. Yet despite these flaws, many sources today assume a low caries experience in the DS population as a fact.[33, 43, 48, 54, 58, 71]

In addition to hypodontia, dental treatment-related factors may also contribute to similar problems in comparing DMFT scores across DS and non-DS groups. Due to the additive characteristic of the DMFT, treatment indicators (MT + FT) are mixed with disease indicators (DT) into one score. One concern about mixing both types of indicators is that dental treatment and prognostic philosophies may vary amongst differing dentists. Also along similar lines, treatment approaches may differ due to a patient's socioeconomic status (with factors such as financial and educational status), and may be due to patient-related factors, such as patients with DS compared to people of the general community.

Bearing these conflicting results and inconsistent study designs in mind, two main studies have demonstrated inconclusive, but noteworthy results. In a prospective study carried out by Vigild,[27] cognitively challenged children were compared with children with DS based on their caries experience. It was found that there was no significant difference in the caries experience among young children with and without DS between the ages of 4 to 12 years, however the caries experience was significantly lower among the older DS cohort between the ages of 13 and 19 years compared to the older control subjects. This study demonstrates the importance of age: it is a significant confounding factor and must always be considered when performing studies on the caries experience.

In addition, in a three-year prospective study involving institutionalized, mentally challenged individuals,[72] the difference between people with and without DS depended on how the caries experience was calculated. It was concluded that when taking into consideration the number of tooth surfaces by using the DMFS index, individuals with DS displayed significantly lower caries. However when the number of teeth were considered by employing the DMFT index, the differences were non-significant. Therefore the method by which the caries experience is calculated must be carefully contemplated in future studies, especially when comparing two groups with very different dental characteristics.

The influence of living arrangements on the caries experience, however, appears to be decisive for people with developmental disabilities, with the exception of people with DS. Despite increases in social acceptance and integration into the community, the oral and dental health of the developmentally challenged is progressively declining. In a study by Gabre and Gahnberg,[3] individuals with mild retardation who lived independently were at highest risk of caries, compared to the severe and moderately challenged. The authors noted that moderate and severe mental retardation was most pronounced among subjects with DS. Among this group with moderate retardation, differences in living arrangement and mental retardation were significant, with an increased caries incidence among the independent compared to institution-dwelling individuals. Despite this finding, the differences between the moderate and severely challenged were non-significant. It was suggested that higher levels of mental retardation impede a person's ability to exercise independent oral care, no matter the living arrangement. With regard to the DS population, the authors argued that dental health is dependent on the decisions made by the caregiver.

On the contrary, other sources have suggested that the increased integration into mainstream society is directly affecting people with DS in a negative manner with respect to dental caries. It was suggested that independent living is conducive to less restricted dietary and oral health regimens compared to the institutionalized living arrangement. Among institutionalized individuals with DS, dieticians are usually present in order to control for their body weight and their tendency towards obesity.[54] These individuals are subject to strict dietary regimens, such as the consumption of less sugar and of more regular meals.

Nonetheless the sociobiological context of people with DS is rapidly changing, with these individuals functioning at a higher level than ever before. The transition from a primarily institution-based, dependent cohort to independent community-dwelling individuals has reflected changes in health, dental and oral risk factors. From a caries research perspective, it will be interesting to capture the balancing act between benefits generated from improved social capital and quality of life, with the potentially negative effects of sustained independent living.

2.5. Summary

DS remains to be the most common cause of genetic intellectual disability among people. In Canada, the prevalence of this disease is increasing while the incidence is expected to remain stable. Although it is known that DS occurs due to the duplication of chromosome 21, its causes are multifactorial but are not concrete. To date, advanced maternal age poses the greatest influence on having a child with DS, where the risk increases exponentially after the age of 35.

People with DS possess numerous health problems, ranging from cardiac to musculoskeletal abnormalities. Dental problems also exist, with many experiencing a high degree of chronic bruxism and periodontitis. However the caries experience of this group remains unclear. Caries research performed on this population in the last 30 years has reproduced conflicting and discordant results. The variability ranges from a low to a high caries experience compared to people without DS. This uncertainty is followed by significant social changes in the challenged community, and by the employment of inconsistent and diverse methods of calculating the caries experience. As such, the aim of this study is to evaluate the caries experience of people with DS compared to those without DS, while taking into account the unique health, physiological and social circumstances of those with DS.

3. OBJECTIVES AND HYPOTHESES

In light of the recent social and health developments among people with DS, additional research on the caries experience of this group must be performed. Therefore the primary objective of this study is to evaluate the caries experience of individuals with DS compared to age- and dentist-matched controls without DS.

It is hypothesized that people with DS will exhibit the same caries experience compared to age- and dentist-matched controls without DS, when taking into account the total number of teeth. However it is anticipated that when the total number of teeth are not being controlled, the DS group will exhibit a lower caries experience compared to the non-DS subjects.

4. METHODOLOGY

This chapter will outline in detail the various steps that were taken in executing this study – from an epidemiological and statistical standpoint.

4.1. Study design

This is a descriptive epidemiologic study that provides information about the caries experience of people with DS. Such a study is a useful tool for examining potential associations, while establishing a "snapshot" of this group's caries experience at a particular period of time.[73]

More specifically this is an observational, matched cross-sectional study where the cases with DS and controls without DS are individually matched based on age and treating dentist. Matching on age aims to control for potential confounding factors that may affect the development of caries from the point of view of environmental exposure. For example, the time of exposure to cariogenic substances, the types of teeth present, and the subject's length of education are factors that are likely to be controlled for. Matching based on the treating dentist ensures that the control subjects are taken from the same "patient pool". As such, matched subjects would be to a certain extent, from similar socioeconomic and cultural backgrounds; while dental treatment and caries diagnosis would also be similar (i.e. restorations would be performed at a similar time).

Subject recruitment of all cases preceded the collection of matched control subjects, and the caries experience for each subject was collected at the same time as the collection of the sociodemographic and clinical data. Each matched group was composed of one subject with DS and two subjects without DS, who are patients of the same dentist and are of the same age (± 1 year) during patient recruitment. Such pairing methodology attempts to increase the statistical power and correspondingly decrease bias – since there is evidence demonstrating that increasing the number of controls to each case improves the accuracy of the results.[74] Ethical approval for this study was granted by the McGill University Institutional Review Board (see appendix 10.17)

4.2. Study population

All case and control subjects were recruited from June 2001 to December 2003. Although the participation rate is unknown, eligible patients were largely present at the dental clinics for routine check-ups.

Each subject followed specific eligibility criteria in order to be included in this study. The cases were derived from a convenience sample from the Montreal Children's Hospital (MCH) dental clinic, and a private dental clinic in Pierrefonds, QC. It was necessary that the cases be composed of individuals with DS, and that the parent or guardian must agree to participate in this study.

Control subjects were individuals without DS. It was necessary that parents or guardians of subjects under sixteen years provide formal permission to participate in this study; while subjects who were sixteen years and older were required to provide their own consent.

In all instances, all individuals who provided formal consent to participate in this study were required to read and complete a consent form in either English or in French. All consent forms were tailored to the location of patient recruitment. (For English-speaking adult subjects, see appendix 10.1 designed for the MCH, and appendix 10.2 designed for the private clinic; and for French-speaking adult subjects, see appendix 10.3 (MCH) and appendix 10.4 (private clinic). For English-speaking parents, see appendix 10.5 (MCH) and 10.6 (private clinic); and for French-speaking parents, see appendix 10.7 (MCH) and 10.8 (private clinic).) In addition, all participants were required to fully comprehend and complete the questionnaires in either French or English.

4.3. Data collection

Information was provided by the treating dentists and by the individuals who provided consent to participate in this study. Data were collected at the dental clinic on the day of patient recruitment.

4.3.1. Dependent variable – Caries experience (Adjusted DFT)

The outcome variable of this study is the caries experience. Traditionally, the DMFT or DMFS scores are used to assess the prevalence of dental caries in an individual. The DMFT score consists of a raw count of the number of decayed, missing and filled teeth due to decay, while the DMFS score is a more detailed assessment representing the number of decayed, missing and filled tooth surfaces due to decay.

In this study, detailed information about each patient's DMFT score and numbers of teeth present at the time of examination were assessed by the treating dentist (see appendix 10.9 for the dental data worksheet). Caries diagnosis was performed through clinical exam only, and was defined at the D_3 level in which decay was detected in dentin with respect to the "D_1-D_3" scale (see appendix 10.10).[75] Subsequent to the data collection, the DMFT score was modified in order to take into consideration hypodontia in the DS group. This mathematical modification helps increase the accuracy of comparing between people with and without DS. The approach to standardize the DMFT score was derived from a study by Ulseth *et al.*[22] Due to potential inaccuracies in determining the causes for missing teeth, the number of decayed (DT) and filled (FT) teeth was divided by the total number of teeth present at the dental examination (see figure 1). Therefore since the caries experience is expressed as a percentage, it is a continuous variable that has a range from 0 to 1.

Figure 1. Adjusted DFT formula

$$\boxed{\text{Adjusted DFT} = \left[\frac{\text{DT} + \text{FT}}{\text{Total \# teeth}} \right]}$$

Two caveats must be raised concerning the comparison of the adjusted DFT scores between the DS and non-DS groups. The first issue relates to the types of teeth involved in this study. Since subject age ranges between 4 and 36 years, there are individuals with either primary or permanent dentition, or a mixture of both. All subjects in the deciduous, mixed and permanent dentition stages were compared together in the analyses, while no differentiation in deciduous or permanent teeth was made in caries experience calculations. Thus to control in a certain extent for the types of teeth present, the case and control subjects were matched by age. The second issue relates to potential differing approaches to caries diagnosis among each participating dentist. Three dentists took part in this study. Thus due to the somewhat subjective nature of the diagnosis of dental caries, calibrations were not implemented to compare every subject's caries experience. However to best control for this factor, this study attempted to match each case and control set by the treating dentist.

4.3.2. Other dependent variables

Although the adjusted DFT is the main dependent variable of interest, other outcome variables that were initially collected by the clinic dentist were also analyzed (see appendix 10.9). These four variables include: the number of decayed, missing, filled teeth and the crude, unadjusted DMFT score.

4.3.3. Independent variables

The second part of data collection involved retrieving independent variables that encompass socio-demographic information, and the clinical dental histories of each subject. This data was provided by the individual who gave consent to participate in this study, by means of filling out a self-completing questionnaire.

Among the cases with DS, parents or guardians were required to fill out an *Oral Assessment in Down syndrome* (OADS) questionnaire (see appendix 10.11 for the English version, and appendix 10.12 for the French version).[76] This tool has been validated in both English and French, and assesses the dental health of people with DS. Specifically, this questionnaire is composed of categorical and Likert-type scales and is designed for caregivers of children with DS.

However in order to compare the cases with the control subjects, eleven questions from the OADS questionnaire that were relevant to the oral health of the non-DS group were extracted for analysis. These same eleven questions were used in a shorter questionnaire for the control group (for English-speaking adult control subjects see appendix 10.13; for French-speaking adult control subjects see appendix 10.14; for English-speaking parents see appendix 10.15; and for French-speaking parents see appendix 10.16.)

4.3.3.1. Sociodemographic variables

Among these eleven items, six questions were devoted to socio-demographic information, where the subject's living arrangement, age, first language, level of education, gender, and ethnicity were assessed. Regarding living arrangement, much literature has supported its effect on the development of caries. For example, individuals who are institutionalized for long periods of time are less likely to experience dental decay compared to those who live at home, due to strict dietary and tooth brushing regimes.[3, 17, 18] This nominal variable is subdivided into five categories, and is only applicable for those respondents with a child with DS.

As for age, it is not only a basic socio-demographic variable, but it also has a profound effect on the caries experience. As a person grows older, the more caries one would accumulate. Since age is a strong confounding factor in this study, this is a key variable for this study's matched design.

In addition, gender is a socio-demographic dichotomous variable that is simple to gather, and it provokes some interest about whether there may be gender differences in the caries experience among people with DS.

With respect to ethnicity, various cultures view dental health differently. For example, the traditional Chinese culture generally does not prioritize personal dental hygiene, compared to people who live in North America for a prolonged period of time.[77] Different cultures possess different diets, which may also affect the caries experience. This is a nominal variable that is subdivided into five ethnic categories that are representative of the demographic composition of Montreal.

Similar to ethnicity, the language of the questionnaire that was completed by the respondent was considered in this study. This is a dichotomous variable subdivided into English and French.

Finally the level of education reflected all subjects who completed the questionnaire. Specifically, subjects who were 16 years and older disclosed their own education level, while the parents or guardians of subjects under 16 years disclosed their own education levels. This ordinal independent variable is subdivided into three levels ranging from elementary school to college to university. Education is often used as a proxy variable for a person's socioeconomic status, as it is related to income and social class. These factors are closely associated with oral health, and also the caries experience.[17]

4.3.2.3. Clinical dental variables

The first question regarding the subject's dental hygiene practices is a nominal variable that asks whether his or her teeth are brushed on a daily basis. This dichotomous variable has an influence on the caries experience, and is used to control for possible differences between the two subject groups.

The subject's clinical dental history was also taken into account. The extent to which bruxism affects the subjects was asked. This is an ordinal variable based on a 4-point scale ranging from 'not at all' to 'very much'. Due to the skewed frequency distribution, this variable was dichotomized into 'yes' and 'no' variables. It is documented that the prevalence of bruxism is high among people with DS and its effects on the caries experience have been widely debated. Some have argued that the smoother surface of the tooth from chronic tooth grinding may inadvertently help decrease a person's chances of developing additional caries.[29, 54, 58, 71, 78]

In addition, two other variables that influence the caries experience is the use of fluoride supplements and exposure to fluoride treatment. These are nominal variables, divided into 'yes', 'no', and 'don't know'. Finally, the presence of any dental problems were initially asked on a 5-point scale ranging from, 'not relevant' to 'at least once a month', was dichotomized into 'yes' and 'no' variables due to the skewed frequency distribution of the sample. This variable is linked to the caries experience, since caries and other oral diseases such as periodontal disease often share similar risk factors, such as oral hygiene, level of cooperation, number of dental care visits, the caregiver's knowledge and attitude towards dental self-care, age, and drug use.[17]

4.4. Statistical analyses

All statistical analyses were performed by using SAS statistical software. Descriptive statistics were used to evaluate the overall composition of both study groups. For each variable measured, frequency distributions, means, proportions and standard deviations were calculated.

With respect to the bivariate and multivariate stages of analysis, two different statistical approaches were made since the dependent variables were all originally continuous. The first approach assumes that the dependent variable is continuous. As such, means and standard deviations were generated. Because the dependent variables did not display normality, non-parametric tests were used for bivariate significance testing. Differences between means in two independent groups were analyzed by the Mann-Whitney test (equivalent to the two-sample unpaired t-test), while the Kruskal-Wallis test (equivalent to the one-way ANOVA) was used to compare several independent groups.

As for the multivariate stage of analysis, multiple linear regression was employed. Since there are five dependent variables, five statistical models were created. However before generating the multivariate models, two verifications for multicausality were conducted to ensure that the results are not biased or misleading. First, a correlation matrix was generated in order to check for potential confounding, where an independent variable may be responsible for at least part of the association between the dependent variable and another independent variable. And second, potential effect modification (or interaction) was determined during this stage of analysis. Finally, in order to verify that multicollinearity (redundancy) was not present in the proposed multivariate models, collinearity diagnostics were performed by analyzing the variance inflation factor (VIF), Eigen values, and proportions of variation.

The second approach assumes that the dependent variable is dichotomous: Subjects were compared with a caries experience of zero versus a caries experience greater than zero, no decayed teeth versus subjects with decayed teeth, no missing teeth due to caries versus subjects with missing teeth, and no filled teeth versus subjects with filled teeth. Specifically among people who did not experience any dental decay, the value for the adjusted and unadjusted DMFT scores are both equal to zero. In this manner, both measures of the caries experience were collapsed together into one category. Therefore four multivariate models were presented in which the odds ratios and 95% confidence intervals were generated for the bivariate analysis stage, and then multiple logistic regression was used for the multivariate analyses. The latter analysis was mainly used to evaluate the relationship between the caries experience and DS status, when controlling for all known predictors of caries.

Before performing the multivariate analyses, tests for multicausality were generated for effect modification (via the Breslow-Day test for homogeneity), and confounding.

With respect to both statistical approaches, all statistical models were controlled for the matched set (in order to maintain the integrity of the 1:2 case/control ratio), dentist, and age due to the matched design of this study. In addition, all variables that contributed to multicausality were eliminated from the multivariate models, and only the significant independent variables found in the bivariate analyses, since they were representative of potential crude associations with the dependent variables. Specifically, those variables that demonstrated a p-value of 0.1 or less, and with confidence intervals that did not cross 1 were included in the overall multiple linear and logistic models. The treatment of the dependent variable as either continuous or dichotomous is beneficial, offering a different "feel" for the data and the results of this study.

5. RESULTS

5.1. Descriptive Analysis

128 subjects were recruited for this study, of which 44 were individuals with DS and 84 were individuals without DS. Table 1 outlines the frequency distribution of all variables measured, subdivided by DS status and by the entire subject population. In terms of the sociodemographic composition of this study, the mean age of the sample was 16.3 years, ranging from 4 to 36 years. The gender distribution was nearly even among both groups, with most subjects living at home with a parent or guardian. There was a near even distribution of subjects subdivided by education level, whereas the majority of the sample was from a Caucasian background. With respect to the place of residence of the subjects, the vast majority of subjects with DS lived at home with a parent or guardian. Although this distribution may be reflective of the age of the study group, this may be indicative of the social change that occurred between the mid-1970s and 1980s in which people with DS moved away from foster institutions and subsequently moved into the general community.

With respect to the dental clinical variables, a large proportion of people with DS experienced various dental problems. For example 61% of all people with DS experienced at least one dental problem in the last year; however this was only apparent in 20% of the non-DS group. Also nearly half the DS sample reported experiencing bruxism, whereas this was only reported among one-third of the non-DS group. The distribution of the use of fluoride supplements differed between the two subject groups. Most people without DS did not take any supplements, whereas 42% of all people with DS reported taking them. Most subjects within both groups underwent fluoride treatment, while a greater majority had their teeth brushed everyday.

The data collected by the dental clinician is also presented in Table 1. The total number of teeth ranged from 14 to 32. Among the controls, the mean number of teeth was 25, with a range from 14 to 32 teeth. Among the cases, no subject had a full set of 32 teeth: the mean number of teeth was 23, ranging from 16 to 30 teeth. In both groups, a large majority of the sample did not have decayed or missing teeth due to caries. Approximately 60% of the DS group did not have fillings, where this was reflected in nearly one-third of the non-DS group. Subjects with DS had a mean DMFT score of 2.6 ranging from 0 to 19, while subjects without DS had a mean DMFT score of 4.9 with a range from 0 to 21. The mean adjusted DFT score was 0.10 among people with DS, which means that 10% of teeth are on average affected by caries. In the same group, approximately 56% of the cases were caries-free. Among people without DS, 18% of teeth were on average affected by caries with approximately 27% of this group having a caries experience of 0.

Table 1. Descriptive statistics for the study sample

VARIABLE	CATEGORY	With DS (n=44) N (%)	Without DS (n=84) N (%)	All subjects (n=128) N (%)
Language	English	24 (54.6)	58 (69.1)	82 (64.1)
	French	20 (45.5)	26 (30.9)	46 (35.9)
Education (respondent)	Elementary/ High school	16 (37.2)	21 (25.0)	37 (29.1)
	College/CEGEP	17 (39.5)	29 (34.5)	46 (36.2)
	University	10 (23.3)	34 (40.5)	44 (34.7)
Age, years (categorized by quartiles for the total sample range)	4-8	12 (27.3)	24 (28.6)	36 (28.1)
	9-12	10 (22.7)	20 (23.8)	30 (23.4)
	13-22	11 (25.0)	20 (23.8)	31 (24.2)
	23-36	11 (25.0)	20 (23.8)	31 (24.2)
Gender	Male	26 (59.1)	39 (46.4)	65 (50.8)
	Female	18 (40.9)	45 (53.6)	63 (49.2)
Ethnicity	White	37 (84.1)	60 (71.4)	97 (75.8)
	Asian	2 (4.6)	11 (13.1)	13 (10.2)
	Other	5 (11.4)	13 (15.5)	18 (14.1)
Residence	With parent/guardian	36 (81.8)	58 (69.1)	94 (73.4)
	Independent	3 (6.8)	26 (31.0)	29 (22.7)
	Communal accommodation	5 (11.2)	0 (0.0)	5 (3.9)
Dental problems	No	17 (38.6)	67 (79.7)	84 (65.6)
	Yes	27 (61.4)	17 (20.2)	44 (34.4)
Bruxism	No	20 (46.5)	59 (70.2)	79 (62.2)
	Yes	23 (53.5)	25 (29.8)	48 (37.8)
Fluoride supplement	No	21 (58.3)	58 (74.4)	79 (69.3)
	Yes	15 (41.7)	20 (25.6)	35 (30.7)
Fluoride treatment	No	7 (22.6)	17 (23.9)	24 (23.5)
	Yes	24 (77.4)	54 (76.1)	78 (76.5)
Daily tooth-brushing	No	2 (4.7)	5 (6.0)	7 (5.6)
	Yes	41 (95.4)	78 (93.9)	119 (94.4)

Table 1 (con't). Descriptive statistics for the study sample

VARIABLE	**CATEGORY**	**With DS (n=44) N (%)**	**Without DS (n=84) N (%)**	**All subjects (n=128) N (%)**
Total number of teeth	14-21	13 (29.6)	11 (13.1)	24 (19.8)
(categorized by quartiles for the total sample range)	22-23	13 (29.6)	12 (14.3)	25 (19.5)
	24-27	11 (25.0)	29 (34.5)	40 (31.3)
	28-32	7 (15.9)	32 (38.1)	39 (30.5)
# Decayed teeth	0	39 (88.6)	65 (77.4)	104 (81.3)
	1-11	5 (11.4)	19 (22.6)	24 (18.8)
# Missing teeth due to caries	0	39 (88.6)	74 (88.1)	113 (88.3)
	1-8	5 (11.4)	10 (11.9)	15 (11.7)
# Filled teeth	0	26 (59.1)	26 (31.0)	52 (40.6)
	1-20	18 (40.9)	58 (69.0)	76 (59.4)
DMFT	0	25 (56.8)	23 (27.4)	48 (37.5)
	1-21	19 (43.2)	61 (72.6)	80 (62.5)
Adjusted DFT	0.00	25 (56.8)	23 (27.4)	48 (37.5)
	0.10-0.63	19 (43.2)	61 (72.6)	80 (62.5)

5.2. Bivariate Analysis

The results of the bivariate analyses of the relationship between the independent variables and each dichotomous dependent variable (adjusted and unadjusted DMFT scores, number of decayed, filled and missing teeth due to caries) are presented in Table 2 (sociodemographic variables) and Table 3 (clinical variables). Table 4 displays sociodemographic variables and Table 5 exhibits clinical variables against all the continuous dependent variables. From two different statistical perspectives, these results show crude estimations of the relationship between the dependent variables and all independent variables in the bivariate models.

A large number of variables have either displayed statistically significant crude associations or a strong tendency toward association between the independent and dependent variables. This is demonstrated in both statistical approaches, when the dependent variable is regarded as either continuous or dichotomous. With respect to all the dichotomous dependent variables, the cut-off points were at zero – a classic categorization in caries-related research. For example with the DMFT outcome, comparisons were made between people with a caries experience (DMFT=0) and people without a caries experience (DMFT>0).

When the sample was divided by DS status, significant differences were found between both groups. Both statistical methods showed that the non-DS group had more filled teeth, and significantly higher adjusted and unadjusted DMFT scores. The odds of having decayed teeth showed a strong tendency toward association (95% confidence interval [CI], 0.8 to 6.6), however it was not statistically significant when measured as a continuous variable (p=0.13). It was interesting to find that there was no statistical difference in the number of missing teeth due to caries between both groups. In general, without controlling for any known predictors for caries, people without DS were roughly 3 times more likely of having a caries experience compared to people with DS, when the DMFT was either unadjusted or adjusted to compensate for the total number of teeth. This difference was also demonstrated when the dependent variables were treated as continuous. The mean differences for the adjusted and unadjusted DMFT scores were highly significant (p<0.001).

Many crude associations were found among the sociodemographic variables. There appeared to be a potential cultural difference in the adjusted and unadjusted DMFT scores, missing teeth due to caries and the number of filled teeth. When English-language speakers were compared with French-language speakers, English speakers were more at risk of having past dental decay, more missing teeth due to caries, and a greater likelihood of having a caries experience – no matter whether the DMFT was adjusted or not. With respect to the number of missing teeth, English speakers were 9.3 times more at risk of having missing teeth due to caries compared to the French speakers (95% CI, 1.2 to 72.2). This difference was also apparent when the mean number of missing teeth were compared (p=0.01). In addition, English speakers were 3.1 times more likely of having a caries experience when the numbers of teeth were considered in the calculation of the DMFT (95% CI, 1.4 to 6.5) and the mean difference between them were statistically significant (p=0.02). From a different perspective, an average of 16% of all teeth among English speakers were affected by caries, compared to 13% of those of French speakers (p=0.06).

The gender and ethnicity of the study subjects and length of education of the respondents did not display significant differences among the caries experience indicators.

As for the subjects' place of residence, there appeared to be a significant difference in the number of missing and filled teeth, and among the adjusted and unadjusted DMFT scores. The relationship between people living independently and those living with a parent or guardian was clear when the dependent variables were dichotomized. Although this may be highly reflective of subject age, the DMFT and adjusted DFT displayed a 4.0 fold risk of a caries experience among independent dwellers compared to those with family (95% CI, 1.4 to 11.5). The p-values were below $\alpha=0.05$ when the number of missing and filled teeth, adjusted DFT and unadjusted DMFT scores were analyzed as continuous. However due to the nature of the Kruskal-Wallis test, it only indicated the presence of a significant difference, and as such could not identify which two categories for place of residence were significantly different.

As for the bivariate analyses for the clinical independent variables, fluoride supplements and treatments, bruxism, the presence of any dental problems, and daily tooth brushing were not significantly associated with any caries experience indicator when the odds ratios were calculated. However with respect to the difference in means, only the crude DMFT and adjusted DFT scores displayed statistically significant differences between people who underwent regular fluoride treatments and those who did not (p=0.03, p=0.01 respectively).

Table 2. Bivariate analyses of dichotomous caries experience indicators with sociodemographic variables (n=128) $^{\Delta}$

	DECAYED (D)			**MISSING (M)**			**FILLED (F)**			**DMFT & ADJUSTED DFT**		
	Decayed /None	**OR**	**95% CI**	**Missing /None**	**OR**	**95% CI**	**Filled /None**	**OR**	**95% CI**	**Caries exp /None**	**OR**	**95% CI**
DS status												
DS	5/39	Ref	Ref	5/39	Ref	Ref	18/26	Ref	Ref	19/25	Ref	Ref
Non-DS	19/65	2.3	0.8-6.6	10/74	1.05	0.3-3.3	58/26	3.2	**1.5-6.9**	61/23	3.5	**1.6-7.5**
Language												
English	17/65	1.5	0.6-3.8	14/68	9.3	**1.2-72.2**	56/26	2.8	**1.3-5.9**	59/23	3.1	**1.4-6.5**
French	7/39	Ref	Ref	1/45	Ref	Ref	20/26	Ref	Ref	21/25	Ref	Ref
Education												
Elementary/HS	10/27	1.7	0.6-4.8	7/30	3.3	0.8-14.0	26/11	2.0	0.8-4.9	27/10	2.1	0.8-5.2
College/CEGEP	6/40	0.7	0.2-2.1	3/43	0.5	0.1-2.4	25/21	1.0	0.4-2.3	27/19	1.4	0.5-2.5
University	8/36	Ref	Ref	5/39	Ref	Ref	24/20	Ref	Ref	25/19	Ref	Ref
Age												
4-8	2/34	Ref	Ref	3/33	Ref	Ref	17/19	Ref	Ref	17/19	Ref	Ref
9-12	7/23	5.2	**1.0-27.1**	2/28	0.79	0.12-5.0	18/12	1.7	0.6-4.5	18/12	1.7	0.6-4.5
13-22	10/21	8.1	**1.6-40.6**	2/29	0.79	0.12-4.8	16/15	1.2	0.5-3.1	19/12	1.8	0.7-4.7
23-36	5/26	3.3	0.6-18.2	8/23	3.8	0.9-16.0	25/6	4.7	**1.5-14.1**	26/5	5.8	**1.8-18.5**
Gender												
Male	9/56	Ref	Ref	8/57	Ref	Ref	35/30	Ref	Ref	38/27	Ref	Ref
Female	15/48	1.9	0.8-4.8	7/56	0.9	0.3-2.6	41/22	1.6	0.8-3.3	42/21	1.4	0.7-2.9

Abbreviation: Ref., reference group for the calculation of odds ratios
OR, odds ratio comparing odds for caries-related indicators within the study sample
Δ Mean numbers refer to the proportion of subjects in each stratified group

Table 2 (con't). Bivariate analyses of dichotomous caries experience indicators with sociodemographic variables (n=128) ∆

	DECAYED (D)			MISSING (M)			FILLED (F)			DMFT & ADJUSTED DFT		
	Decayed /None	**OR**	**95% CI**	**Missing /None**	**OR**	**95% CI**	**Filled /None**	**OR**	**95% CI**	**Caries exp /None**	**OR**	**95% CI**
Residence												
With parent/guardian	17/77	Ref	Ref	7/87	Ref	Ref	49/45	Ref	Ref	51/43	Ref	Ref
Independent	5/24	0.9	0.3-2.8	7/22	4.0	**1.3-12.5**	23/6	3.5	**1.3-9.4**	24/5	4.0	**1.4-11.5**
Communal Accommodation	2/3	3.0	0.5-19.5	1/4	3.1	0.3-31.7	4/1	3.7	0.4-34.1	5/0	--	--
Ethnicity												
White	16/81	2.0	0.6-6.2	12/85	0.9	0.2-4.3	54/43	1.3	0.5-3.5	56/41	1.5	0.5-4.2
Asian	3/10	1.3	0.3-6.7	1/12	1.5	0.1-18.5	11/2	0.3	0.5-3.5	12/1	0.2	0.1-1.6
Other	5/13	Ref	Ref	2/16	Ref	Ref	11/7	Ref	Ref	12/6	Ref	Ref

Abbreviation: Ref., reference group for the calculation of odds ratios
OR, odds ratio comparing odds for caries-related indicators within the study sample
∆ Mean numbers refer to the proportion of subjects in each stratified group

Table 3. Bivariate analyses of dichotomous caries experience indicators with clinical independent variables (n=128) $^{\Delta}$

	DECAYED (D)			MISSING (M)			FILLED (F)			DMFT & Adjusted DFT		
	Decayed /None	**OR**	**95% CI**	**Missing /None**	**OR**	**95% CI**	**Filled /None**	**OR**	**95% CI**	**Caries exp /None**	**OR**	**95% CI**
Dental problems												
No	15/69	0.8	0.3-2.1	7/77	0.4	0.1-1.2	51/33	1.2	0.6-2.5	53/31	1.1	0.5-2.3
Yes	9/35	Ref	Ref	8/36	Ref	Ref	25/19	Ref	Ref	27/17	Ref	Ref
Bruxism												
No	16/63	1.3	0.5-3.2	11/68	1.8	0.5-5.9	49/30	1.3	0.7-2.9	52/27	1.5	0.7-3.1
Yes	8/40	Ref	Ref	4/44	Ref	Ref	26/22	Ref	Ref	27/21	Ref	Ref
Fluoride Supplements												
No	19/60	Ref	Ref	9/70	Ref	Ref	46/33	Ref	Ref	48/31	Ref	Ref
Yes	4/31	0.4	0.1-1.3	5/30	1.3	0.4-4.2	24/11	1.6	0.7-3.6	26/9	1.9	0.8-4.5
Fluoride Treatments												
No	7/17	2.3	0.8-6.6	3/21	1.3	0.3-5.1	16/8	1.5	0.6-3.8	18/6	2.1	0.7-5.9
Yes	12/66	Ref	Ref	8/70	Ref	Ref	45/33	Ref	Ref	46/32	Ref	Ref
Daily tooth-brushing												
No	2/5	1.9	0.3-10.3	2/5	3.2	0.6-18.5	5/2	1.7	0.3-9.4	5/2	1.6	0.3-8.5
Yes	21/98	Ref	Ref	13/106	Ref	Ref	70/49	Ref	Ref	73/46	Ref	Ref

Abbreviation: Ref., reference group for the calculation of odds ratios
OR, odds ratio comparing odds for caries-related indicators within the study sample
Δ Mean numbers refer to the proportion of subjects in each stratified group

Table 4. Bivariate analyses of continuous caries experience indicators with sociodemographic variables (n=128)

	DECAYED (D)		**MISSING (M)**		**FILLED (F)**		**DMFT**		**ADJUSTED DFT**	
	Mean (SD)	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**
DS status*										
DS	0.25 (0.78)		0.32 (1.12)		2.02 (3.35)		2.59 (4.20)		0.10 (0.15)	
Non-DS	0.63 (1.68)	0.13	0.33 (1.18)	0.93	4.30 (4.63)	**0.00**	4.90 (4.94)	**0.00**	0.18 (0.18)	**0.00**
Language*										
English	0.50 (1.23)		0.49 (1.40)		4.01 (4.56)		4.63 (4.93)		0.16 (0.17)	
French	0.50 (1.77)	0.45	0.04 (0.29)	**0.01**	2.63 (3.84)	**0.03**	3.17 (4.49)	**0.02**	0.13 (0.19)	0.06
Education**										
Elementary/HS	0.70 (1.94)		0.41 (1.01)		4.51 (4.74)		5.62 (5.68)		0.19 (0.19)	
College/CEGEP	0.41 (1.33)		0.22 (1.19)		3.30 (4.65)		3.93 (5.28)		0.14 (0.18)	
University	0.43 (1.07)	0.34	0.39 (1.24)	0.24	2.91 (3.65)	0.21	3.73 (4.40)	0.23	0.13 (0.17)	0.23
Age**										
4-8	0.06 (0.23)		0.11 (0.40)		0.47 (0.51)		2.56 (3.69)		0.11 (0.16)	
9-12	0.23 (0.43)		0.20 (0.81)		0.60 (0.50)		3.73 (4.34)		0.16 (0.20)	
13-22	0.32 (0.48)		0.06 (0.25)		0.52 (0.51)		3.74 (4.94)		0.13 (0.18)	
23-36	0.16 (0.37)	0.07	0.97 (2.04)	**0.03**	0.81 (0.40)	**0.00**	7.60 (6.09)	**0.00**	0.21 (0.18)	**0.03**
Gender*										
Male	0.46 (1.35)		0.32 (1.26)		3.52 (4.82)		4.06 (5.02)		0.15 (0.19)	
Female	0.54 (1.54)	0.23	0.33 (1.03)	0.90	3.51 (3.86)	0.45	4.16 (4.63)	0.64	0.15 (0.18)	0.59

* Mann-Whitney U test
** Kruskal-Wallis test

Table 4 (con't). Bivariate analyses of continuous caries experience indicators with sociodemographic variables (n=128)

	DECAYED (D)		**MISSING (M)**		**FILLED (F)**		**DMFT**		**ADJUSTED DFT**	
	Mean (SD)	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**
Residence**										
With parent/guardian	0.38 (1.29)		0.16 (0.66)		2.71 (3.69)		3.26 (4.40)		0.13 (0.18)	
Independent	0.76 (1.81)		0.72 (1.77)		5.72 (5.50)		6.17 (5.41)		0.19 (0.17)	
Communal accommodation	1.20 (1.79)	0.38	1.20 (0.68)	**0.04**	5.80 (4.09)	**0.00**	8.20 (3.27)	**0.00**	0.30 (0.14)	0.11
Ethnicity**										
White	0.35 (0.89)		0.39 (1.30)		3.45 (4.40)		4.20 (5.22)		0.14 (0.18)	
Asian	0.69 (1.93)		0.08 (0.28)		4.08 (3.86)		4.84 (3.74)		0.21 (0.19)	
Other	1.17 (2.79)	0.44	0.17 (0.51)	0.84	3.44 (4.62)	0.49	4.78 (0.39)	0.39	0.16 (0.19)	0.20

* Mann-Whitney U test
** Kruskal-Wallis test

Table 5. Bivariate analyses of continuous caries experience indicators with clinical independent variables (n=128)

	DECAYED (D)		MISSING (M)		FILLED (F)		DMFT		ADJUSTED DFT	
	Mean (SD)	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**	**Mean (SD)**	**p**
Dental problems*										
No	0.48 (1.49)		0.20 (0.78)		3.42 (4.24)		4.10 (4.90)		0.15 (0.18)	
Yes	0.55 (1.35)	0.68	0.57 (1.62)	0.10	3.70 (4.60)	0.94	4.82 (5.56)	0.63	0.17 (0.19)	0.67
Bruxism*										
No	0.56 (1.65)		0.41 (1.32)		3.35 (4.28)		4.32 (5.07)		0.14 (0.18)	
Yes	0.42 (1.05)	0.68	0.21 (0.82)	0.34	3.63 (4.42)	1.00	4.25 (5.22)	0.65	0.16 (0.20)	0.92
Fluoride Supplements*										
No	0.56 (1.26)		0.39 (1.35)		3.60 (4.77)		4.54 (5.61)		0.16 (0.19)	
Yes	0.49 (1.96)	0.15	0.20 (0.53)	0.75	3.74 (3.65)	0.39	4.43 (4.27)	0.49	0.17 (0.16)	0.38
Fluoride Treatments*										
No	0.88 (1.85)		0.17 (0.48)		5.00 (5.48)		6.00 (5.37)		0.24 (0.22)	
Yes	0.32 (0.85)	0.12	0.38 (1.36)	0.85	3.03 (3.69)	0.16	3.37 (4.11)	**0.03**	0.12 (0.15)	**0.01**
Daily tooth-brushing *										
No	0.43 (0.79)		0.43 (0.78)		5.57 (6.75)		6.29 (6.42)		0.24 (0.24)	
Yes	0.48 (1.45)	0.54	0.33 (1.18)	0.19	3.36 (4.15)	0.41	3.92 (4.07)	0.33	0.15 (0.17)	0.34

* Mann-Whitney U test
** Kruskal-Wallis test

5.3. Multivariate Analysis: multiple logistic regression

5.3.1. Tests for multicausality

For each dependent variable, crude odds ratios (ORs) for DS status were compared against various adjusted ORs for DS status (where independent variables were added one by one to the model), in order to detect confounding. By convention, if the ORs increased by over 10%, the independent variable that was added last to the respected model may be considered as a confounder.[79] In this study for each dependent variable, the ORs for DS status remained stable no matter whether variables were added to the models. As such, none of the multivariate logistic models exhibited any confounding.

As for verifying effect modification, it was believed that there may be a potential interaction between DS status, fluoride treatment and the caries experience (adjusted and unadjusted DMFT). Specifically, the effect of fluoride treatment on the caries experience was thought to change between people with and without DS. A Breslow-Day test for effect modification was performed, however this phenomenon was not significant (p=0.19).

After conducting the two tests for multicausality, no variables were found to potentially produce biased or unreliable results. As such, only those independent variables that were found to be statistically significant at the bivariate stage of analysis were included in each multiple logistic regression model.

5.3.2. Multivariate models

The results of the four multiple logistic regression models are shown in Table 6. With respect to the caries experience (as measured by either the adjusted or unadjusted DMFT scores), age, language and fluoride treatment were found to be significant in the bivariate stage of analysis. Therefore, these variables were incorporated into the multivariate model. As such, it was found that subjects without DS had a 4.58 greater risk of having a caries experience (with an adjusted or unadjusted DMFT greater than 0) compared to subjects with DS when controlling for all other known predictors (95% CI, 1.49 to 14.08). As for age, every year increase corresponds with an increased likelihood of having a caries experience by a factor of 1.16 (95% CI, 1.06 to 1.27) when controlled for all other known predictors in the model. From a broader perspective, a five year change in age will increase the risk of having a caries experience by a factor of 2.1. Fluoride treatment and language, however, did not display any statistical significance with respect to the caries experience.

With respect to the presence of decayed teeth, only age showed to be significant in the bivariate stage of analysis. As expected, this variable did not reflect any statistical significance in this model, nor were there significant differences found between people with and without DS.

As for the presence of missing teeth due to dental decay, age and language were significant in the bivariate stage of analysis. When they were added to the multivariate model, only age appeared to be statistically significant. It showed that for each increase of one year in age, the likelihood of having missing teeth due to caries increases by a factor of 1.06 (95% CI, 1.00 to 1.12).

With the presence of filled teeth as a dependent variable, age and language were significant at the bivariate stage of analysis. In this particular multivariate model, it was found that subjects without DS were 3.82 times more likely of having filled teeth compared to subjects with DS, when controlling for age and language (95% CI, 1.62 to 9.00). Age was also significant in that as a person gets older each year, the risk of having filled teeth is 1.06 when controlled for all variables in the model (95% CI, 1.01 to 1.12).

5.4. Multivariate Analysis: multiple linear regression

5.4.1. Tests for multicausality and multicollinearity

With respect to confounding, five separate correlation matrices were generated against all independent variables used in this study. In order to detect confounding, there must be a sufficiently high correlation (by convention, values above ±0.4) between two risk factors, while these particular variables must be highly correlated to the outcome.[80] However among each of the five models, this phenomenon did not occur. As such, there was no evidence of confounding.

Effect modification was tested among DS status, fluoride treatment and the caries experience (adjusted and unadjusted DMFT). (Please refer to section 5.3.2. for the rationale). When comparing between the two statistical models with and without the interaction term (fluoride treatment * DS status), the parameter estimate for the caries experience did not significantly change, nor was the interaction term significant (p=0.12 for the adjusted DFT, and p=0.18 for the unadjusted DMFT). As such, effect modification was not present in the proposed multivariate models.

In order to detect multicollinearity (or redundancy) in each multivariate model, proportions of variation, VIF and Eigen values were calculated. All tests did not show the presence of multicollinearity: Within each model, all VIF values were approximately equal to 1 (where by convention, values over 3 may be considered as collinear), all Eigen values fell below 3 (values over 100 generally reflect collinearity), and all proportions of variation were under 0.7 (values greater than 0.7 generally reflect collinearity).[80]

Therefore since no variables were found to potentially generate biased or unreliable results, only those independent variables that were found to be statistically significant in the bivariate stage of analysis were included in the particular multiple linear regression models.

5.4.2. Multivariate models

In this stage of analysis, the dependent variables were continuous, against independent variables that were initially found to be statistically significant during bivariate analyses. Table 7 displays the results of the five multiple linear regression models.

With respect to the adjusted DFT, when adjusted for age, dentist, matched set, language, and fluoride treatment, people with DS had on average 5% fewer teeth affected by caries, compared to subjects without DS. This difference, however, was not statistically significant (p=0.18). On the other hand, age was found to be a significant factor in the same model. It was found that as age increases by one year, an average of 1% of all teeth are affected by caries when controlled for all other variables in the model (p=0.03). Alternatively, one can interpret that every 10 years, an average of 10% of all teeth are affected by caries, when controlled for all other variables in the model.

The DMFT score was observed to be borderline significant with DS status when controlling for age, dentist, matched set, language, and fluoride treatment (p=0.06). This demonstrated that subjects with DS had on average 1.77 fewer teeth affected by caries compared to subjects without DS. Age was also found to be highly significant, in which an average of 0.22 teeth are affected by caries as age increases by one year when controlling for all known predictors in the model (p<0.001).

As for the number of decayed teeth, DS status was controlled for age, dentist, and matched set. This model showed no significant differences between people with and without DS with respect to the number teeth affected by dental decay present at the time of examination.

The number of missing teeth due to caries did not show any significant difference between subjects with and without DS, when controlled for age, dentist, matched set, and language (p=0.95). Age was the only significant variable demonstrating that an average of 0.03 teeth are missing as one gets older each year (p=0.00).

Finally the number of filled teeth demonstrated a significant difference between both DS groups. When controlled for age, dentist, matched set, and language, people without DS had on average 2.29 more filled teeth than people with DS (p<0.001). Age also showed an expected significant difference within the same MLR model, in which an average of 0.13 teeth are filled as age increases by one year (p<0.001).

Table 6. Multiple logistic regression analysis of the relationship between DS status and various caries experience indicators

Variables	**Categories**	**Decayed teeth**		**Missing teeth**		**Filled teeth**		**DMFT & Adjusted DFT**	
		Odds ratio	**95% CI**	**Odds ratio**	**95% CI**	**Odds ratio**	**95% CI**	**Odds ratio**	**95% CI**
DS status	DS	Ref	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Non-DS	2.45	0.81 to 7.42	0.75	0.22 to 2.63	3.82	**1.62 to 9.00**	4.58	**1.49 to 14.08**
Age	Continuous variable	1.04	0.98 to 1.10	1.06	**1.00 to 1.12**	1.06	**1.01 to 1.12**	1.16	**1.06 to 1.27**
Language	English	--	--	7.33	0.87 to 61.72	2.02	0.89 to 4.60	1.95	0.70 to 5.42
	French	--	--	Ref	Ref	Ref	Ref	Ref	Ref
Fluoride treatment	No	--	--	--	--	--	--	Ref	Ref
	Yes	--	--	--	--	--	--	0.89	0.22 to 3.70

Abbreviation: Ref., reference group for the calculation of odds ratios

*In order to maintain the integrity of this matched study, all multivariate models were also controlled for the matched set and treating dentist

Table 7. Multiple linear regression analysis of the relationship between DS status and various caries experience indicators

Variable	**DECAYED TEETH**		**MISSING TEETH**		**FILLED TEETH**		**DMFT**		**ADJUSTED DFT**	
	Parameter estimate	**P**	**Parameter estimate**	**P**	**Parameter estimate**	**P**	**Parameter estimate**	**P**	**Parameter estimate**	**P**
DS status	-0.38	0.14	0.01	0.95	-2.29	**0.00**	-1.77	**0.06**	-0.05	0.18
Age	0.02	0.09	0.03	**0.00**	0.13	**0.00**	0.22	**0.00**	0.01	**0.03**
Language	--	--	-0.31	0.14	-0.33	0.67	-0.38	0.70	0.01	0.88
Fluoride treatment	--	--	--	--	--	--	-1.54	0.19	-0.11	**0.01**

*In order to maintain the integrity of this matched study, all multivariate models were also controlled for the matched set and dentist

6. DISCUSSION

The overall objective of this study was to evaluate the caries experience among people with DS, when compared to people without DS. Two general conclusions can be generated from this study: 1) The caries experience, when expressed as a percentage of caries-affected teeth, is the same among people with and without DS, and 2) The caries experience, when the total numbers of teeth are not taken into account, is lower among people with DS compared to people without DS.

The aforementioned results differ with respect to the consideration of hypodontia and delayed eruption in the calculation of the caries experience. Nevertheless, both approaches and results are consistent with a number of studies involving people with DS. Only two studies have overtly controlled for the numbers of teeth in caries experience calculations, and can attest to the first conclusion where the prevalence of dental decay is not significantly different among people with and without DS. The first paper by Ulseth *et al.* demonstrated the caries experience as a percentage of decayed and filled teeth, while matching for gender, age, level of functioning and medication use.[22] This study did not include the number of missing teeth due to decay in caries experience computations due to the possibility of error in deciphering causes of missing teeth among the subjects. Nevertheless no statistically significant differences were found in the caries experience among people with DS and cognitively challenged individuals living in institutions.

The second paper by Cutress was based on a large-scale study involving over 1,500 subjects.[19] The author presented the DMFT score as a percentage of caries-affected teeth (%DMFT). It was found that there were no significant differences between the DS, cognitively challenged and 'normal' groups, after controlling for living arrangement and age.

The second conclusion, in which the caries experience is significantly lower among the DS group compared to the non-DS group when the total numbers of teeth are not taken into account, is supported by many other studies.[2-17, 19] This is an important issue, and will be discussed in section 6.2.1 regarding methodological inconsistencies when comparing the caries experience of DS and non-DS populations.

Since the caries experience is not concrete among the DS population, a multitude of hypotheses have been generated amongst all 60 studies pertaining to this cohort. However after reflecting on the results of this study and those of past studies, conflicting results concerning the caries experience of people with DS may be reduced to three major factors. First, there may be a possibility that the environment may play an important role in influencing the caries experience; second, biological differences between people with and without DS may elicit varying results; and third, dental treatments may considerably vary between both groups.

6.1. Environmental factors and the caries experience

Biological and physiological factors may not be the sole driving causes for differing caries experiences between the DS and general populations. Where an individual resides may be an important predictor for dental caries. When comparing between people living at home with those in institutions, much evidence has supported poor oral health among the institutionalized.[81, 82] However with respect to people with DS and other cognitive disabilities such as cerebral palsy, some studies have demonstrated the opposite effect. It has been reported that the caries experience is significantly lower among the challenged living in institutions, compared to the same group of people living in the general community.[18-20, 22] Furthermore, there is evidence that the caries experience may be no different among the challenged living in the community and 'normal' people of the general population.[19, 20]

A study by Cutress can best demonstrate these phenomena.[19] This sample included DS, disabled, and 'normal' subjects, and were subsequently subdivided by living arrangement. Among one of the author's analyses, subjects who lived at home in the community were compared with subjects who lived in institutions. It was found that the caries experience was the same among all institutionalized subjects with DS and other disabilities. For example in the 20-24 age group, the mean DMFT was 9.2 for DS subjects and 6.4 for the disabled. This difference was not statistically significant. However, when compared to all other subjects who lived in the general community, the caries experience was significantly higher – irrespective of disability. All DS, disabled, and 'normal' subjects had high DMFT scores of approximately 17: a value significantly higher than all institutionalized subjects in the sample.

With respect to this current study, the results were predominantly reflective of people with DS living at home with their family. (Only 11% of all subjects with DS reported to live in institutions.) From these data, a similar trend was achieved. It was found that the caries experience was the same among the non-DS group and community-dwelling subjects with DS – inferences that closely mirror the results of Cutress, Kroll, and Nowak.[18-20]

Therefore given the results of this study and that of past studies, having DS may not have a unilateral effect on the caries experience. Instead, the living arrangement or environment of a person with DS may be very important contributors that are worth investigating in future caries-related studies. Since there is some evidence of differing caries experience with respect to living arrangement – no matter whether a person has DS or not – the reasons that closely affect oral health appear to be multifactorial. It may be suggested that such factors could possibly revolve around caregivers' access to resources and behaviours surrounding oral health care. For people with DS living at home, dental care may not be as readily available to caregivers – an important resource that was often provided in institutions.[83-84] Dental self-care practices are therefore reported to be less controlled in community settings,[46] and such individuals are not only subject to more caries, but poorer oral hygiene and a higher risk of periodontal disease.[85]

6.1.1. *Physical access to a dental professional*

For many families with a challenged child such as DS, it is often very difficult to access a dental professional. The distance may be too far: A study in California demonstrated that the distance travelled by special-care patients have increased in the late 1980s compared to the late 1970s.[86] Alternatively, many cannot get a ride to the dentist: People with disabilities were nearly 10 times more likely to not attend regular dental check-ups because of the lack of transportation, compared to non-disabled patients.[87]

In addition, there are not enough trained dentists in the special-care field, and the number of general dental professionals willing to treat people with disabilities is deficient. Approximately 20% of all dental professionals in the United States treat patients with disabilities.[46] In France, it is reported that people with DS are nearly two-times more likely to have difficulty finding a dentist compared to finding a physician.[45] However among those who treat the underprivileged, a large majority do not have any advanced training or sufficient experience in this field.[46]

6.1.2. *Financial access to a dental professional*

To many, dental care is costly, especially for people caring for individuals with numerous health and developmental problems. The inability to pay and the lack of dental insurance are significant factors in deterring caregivers from giving their developmental child proper dental maintenance and treatment.[46] However in institutions, dental services are not only readily available, but the costs are often covered by the institutions themselves.

6.1.3. *Caregiver behaviour towards oral health care*

Oral health care is often considered a secondary priority to caregivers of children with DS, especially when they are prone to many serious health problems (i.e. cardiac, ENT, immunological). It can be inferred from a study by Allison *et al.* that the importance of dental self-care is not as high among people with DS, compared to their siblings.[45] For example, teeth are not as likely to be brushed especially at a younger age for those with DS compared to their non-DS siblings. Also the odds of having dental treatment of any kind were highest among the non-DS group across all age groups.[45]

In addition, diets are often poor in energy and nutrients among people with DS living at home.[88] In the institutions, the environment is highly-controlled with the presence of trained dieticians in the field of special-care. Foods are usually lower in calories in order to regulate above-average body mass index (BMI) levels,[89] and are consumed at regular time intervals compared to the home setting.[54] As such, the importance or motivation to maintain oral hygiene may be lacking amongst caregivers or independent individuals, promoting a negative effect on the oral health of people with disabilities.

6.2. Host-related factors and the caries experience

6.2.1. *Congenitally missing teeth*

It is widely recognized that people with DS generally have fewer teeth compared to people without DS. The total number of teeth must be taken into account when comparing the caries experience with other groups of people. Avoiding this significant physiological factor brings about many biases in the results. As already mentioned, only two studies demonstrated the caries experience as a percentage of caries-affected teeth, and both demonstrated non-significant differences among the two populations, when controlled for age and living arrangement.[19, 22] The adjustment of the DMFT score by expressing the prevalence of dental decay as a proportion of caries-affected teeth avoids the underestimation of the caries experience, and is a good relative measure of disease severity. This method therefore increases the accuracy of comparing the two groups due to the differences in the number of teeth.

To further demonstrate that people with DS have fewer teeth, a high number of missing teeth was found among the DS group in this study. Among the cases, an average of 2.1 teeth were missing due to hypodontia or oligodontia, disease or delayed eruption, with 52% of this group having lost at least one tooth. The controls had on average 1.0 missing tooth, with 31% of that group missing at least one tooth. This large difference between both groups is a testament that all teeth present must be considered for caries experience calculations.

A simple count of the number of decayed, missing and filled teeth may often be misleading when comparing individuals with different numbers of teeth in the mouth. For example in this study when controlled for all predictors of caries, people without DS were nearly 5 times more likely to have a caries experience when the numbers of teeth were not factored into the calculation (95% CI, 1.5 to 14.0). From a different perspective, people without DS had an average DMFT of 1.8 higher than to people with DS (p=0.06). However when the DFT was expressed as a percentage of teeth affected by caries, there was no longer a statistical difference between the two groups (p=0.18). As such, partial edentulism due to delayed eruption or congenital reasons may be a major factor in the discrepant results of past studies, demonstrating a low caries experience among the DS group.

6.2.2. *Prior hypotheses*

Provided the inconsistent research methods on studies involving the caries experience of people with DS, one must thus be vigilant when assessing arguments surrounding this epidemiological debate. Much literature speculated that the low prevalence of decay among this population was primarily due to the physiological differences among them. Emphasis was placed on the number of *Streptococcus mutans* and salivary pH; however more current research has led these hypotheses to be inconclusive.

For example the bacteria, *Streptococcus mutans* in plaque and saliva are partially responsible for the development of dental caries. Some studies have suggested that the concentrations of *S. mutans* among individuals with DS are significantly lower than the normal population.[58] However later studies have been vigilant about making such conclusions, as it was found that there were no differences in *S. mutans* counts among preadolescent and adult DS populations, versus healthy and non-DS subjects with cognitive challenges.[5, 13]

In addition, many older studies have suggested that parotid gland metabolism among individuals with DS is altered due to excess bicarbonate in the saliva, increasing its pH level.[58, 71, 78, 90] It is widely believed that this increase in salivary pH helps buffer cariogenic acids in the mouth, promoting a decreased risk of caries. However more recent research has found that the relationship between salivary pH and caries levels is inconclusive. Studies by Stabholz *et al.* and Shapira *et al.* have not found any significant differences between these two variables among children and adults with DS, and the general population.[5, 13] And to further deflate this proposition, a very recent study has reported an opposite result such that salivary pH is lower compared to non-DS controls.[91]

6.3. Dental treatment approaches and the caries experience

Due to varying differences in anatomy and behaviour, the level of basic dental care among people with DS differs compared to the general population.[1] The level of cooperation among the DS population varies with respect to the level of fear and anxiety, neuromuscular limitations, emotional status and cognitive function.[46] Because of these difficulties, the dentists' approaches to preventative treatments and treatments of diseases such as dental caries may be compromised – giving rise to differing caries experiences among the DS and non-DS populations. Nevertheless, the treatment approach should be equitable among both groups. A study by Shapira and Stabholz demonstrated a successful 30-month preventive dental health program among people with DS.[29] Overall plaque and gingival indices decreased, while the caries experience (as expressed as the number of affected surfaces per child) also decreased significantly. As such, the authors urged for a multidisciplinary network of dental professionals to help alleviate dental problems in the DS population.

6.4. Study limitations and future directions

It must be recognized that there were several limitations in this study that may compromise the generalizability and applicability of the results.

6.4.1. *Sample size*

The overall sample size of this study was small (n=128). In order to maximize statistical power and thus be able to reliably detect important differences between both groups, the sample size must be increased via proper sample size calculations.[73] Many potential associations may have been missed, given that some variables showed very strong tendencies toward significance; one such example is the comparison between the cases and controls with respect to the unadjusted DMFT score (p=0.06). As such, a larger sample size would better highlight the true associations between the unadjusted DMFT score and DS status in the multivariate model, and further reinforce the similarities between both groups when the caries experience is expressed as a percentage of teeth affected by caries (adjusted DFT score). In addition, there is a need to increase the sample size of the DS group living in institutions. This is to better control for the effects of living arrangement on the caries experience, and to clarify differences amongst them and community-dwellers in a 21st century social context.

6.4.2. *DS classifications*

There are three genetic forms of DS. Because genetics is one of multiple of factors that contribute to the caries experience, future studies may subdivide subject groups by the three DS genotypes. Each group may exhibit different dental aetiologies and possibly different caries experiences. Although the feasibility of such study may be low since approximately 94% of all cases with DS concern Standard Trisomy 21 (non-disjunction), it would be interesting to see if clinical differences exist among them.

6.4.3. *Statistics*

There is a limitation with respect to the statistics used in this study. Multiple logistic regression was mainly used to demonstrate how the caries experience is greater among the non-DS group compared to the subjects with DS, without taking into account the total number of teeth. This methodology is correct for any type of variable, as long as it is dichotomized.

However difficulties were encountered with the multiple linear regression approach, in order to evaluate the degree of the caries experience between both study groups. The dependent variables, with the exception of the adjusted DFT, are all integers. Although they may be considered as continuous variables, the issue lies in the large number of zeros in the data. Multiple linear regression is generally tailored for numbers greater than zero. Logarithmic transformations did not alleviate the underlying distributional assumptions (linearity, homoscedasticity, and normality). As such these assumptions were unfortunately violated in the number of decayed, missing, filled teeth, and the unadjusted DMFT score.

In order to take these factors into consideration, two debatable statistical approaches can be used to alleviate this problem. First, non-parametric multiple regression techniques can be used to mainly deal with the non-normal data. This is a very complex procedure, and is rarely used in epidemiological studies. Second, Conditional Poisson Regression (CPR) may be used. This is a novel statistical technique that explicitly deals with extra zeros in the dataset, and when the data are integers.[92] The CPR method compromises of two parts: a) A standard, multiple logistic regression model in which data are compared when the dependent is zero (i.e. no caries experience), versus data greater than zero (i.e. caries experience present), and b) A Truncated Poisson Regression model in which data are analyzed given that the data are greater than zero. In the case of this study, only people who have a caries experience would be analyzed, and then compared between people with and without DS. Many statisticians may argue against this method since the Truncated Poisson Regression model is based on the assumption that the data leads to infinity. This study has an upper-limit of 32: there are normally a maximum of 32 teeth that can be affected by caries. In short, there is no one solution to deal with this study's dataset.

6.4.4. *Method of subject recruitment*

There were notable imperfections in the sampling method of this study. All subjects were derived from a convenience sample from two dental clinics and as such, the results are not truly reflective of the general DS population. Compared to those who do not regularly see a dentist, it may be likely that those patients who seek regular dental care may be of different oral health, from a higher socioeconomic status, or be more informed of the importance of oral care regardless of the many health problems one may face. Therefore in order to make these results generalizable and to avoid sampling bias, it is vital that sampling be randomized across various regions and cultures. Nevertheless, numerous other factors that may influence the caries experience were taken into consideration in this study. The matching methodology by age and dentist has helped adjust for many factors that could not otherwise be overtly controlled for in the study.

6.4.5. *Causes for missing teeth*

Not only were there limitations in the sample source, but the causes for missing teeth were on occasion unclear. As in all calculations of the DMFT score, the ‘missing teeth’ variable (MT) must only be due to dental decay. Due to this uncertainty, the numbers of missing teeth were excluded from the adjusted caries experience indicator (adjusted DFT). Future studies should include missing teeth due to decay. Thus to improve the accuracy of caries experience calculations, the number of decayed, missing, and filled teeth should be divided by the number of decayed, missing, filled, and sound teeth in each subject.

6.4.6. *Caries experience evaluations*

As previously mentioned, it is best to express the DMFT score as a percentage of caries-affected teeth when comparing two different groups with varying numbers of teeth. However it would be also interesting if future studies also employed the DMFS score divided by the total number of tooth surfaces as another measure of the caries experience, to further strengthen (or possibly weaken) the current debate on individuals with DS. Yet when using either a DMFT or DMFS score, calibrations for caries diagnosis must be made a priority in future studies. Caries diagnosis agreements were not assessed in this particular study, and due to its subjective nature, all involved clinicians must have similar methods and judgments in determining teeth that have been affected by decay.

6.5. Clinical implications

The aetiology of caries among the DS population elicits added complex factors compared to the general population. People with DS require extensive oral health needs, not only to improve oral hygiene or to decrease the likelihood of periodontal disease, but it is also to help decrease the caries experience. Given the results of this study, and of other supporting studies, it is suggested that caregivers and dental professionals should not overlook the severity of dental decay among their patients with DS.

7. CONCLUSIONS

While recognizing the limitations of this study, the results suggest that the proportional caries experience is the same among people with and without DS, when controlling for all known predictors.

Peripheral conclusions:

- When the total numbers of teeth are not taken into account in the calculation of the caries experience, people without DS were nearly 5 times more likely of having a caries experience compared to people with DS.

- When the total numbers of teeth are not taken into account in the calculation of the caries experience, people without DS had on average 1.8 more teeth affected by caries compared to people with DS.

- Due to the dependency of tooth count in the calculation of the DMFT, all teeth should be considered when making comparisons of the caries experience between people with and without DS.

- The adjusted DFT score, when expressed as a percentage of caries-affected teeth, is a relative gauge of disease severity. This is a more accurate measure when comparing different groups with different numbers of teeth.

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10. APPENDIX

10.1. English consent form (MCH) for adult subjects

DEVELOPMENT OF A QUESTIONNAIRE TO ASSESS ORAL HEALTH PROBLEMS IN PEOPLE WITH DOWN SYNDROME

Dr. P.J. Allison
Faculty of Dentistry, McGill University

CONSENT FORM

Introduction
We are interested in finding out about the oral and dental health problems experienced by people with Down syndrome in Canada. There are currently no data concerning the dental health and the dental care provision for people with Down syndrome in this country. However, evidence from other countries suggests that people with Down syndrome have a high level of oral and dental disease but have major problems accessing dental services. In order to assess the extent of oral problems, this study requires that we seek those individuals with and without Down syndrome in order to compare the two groups.

If you agree to participate, what will that involve?
PARTICIPATION IS ENTIRELY VOLUNTARY. If you agree to participate, all that would be required is for you to complete our questionnaire with respect to yourself and allow the clinic dentist (Dr. Schwartz) to examine you, and collect data for the study that she often routinely collects anyway. Such data would include the number of fillings you may have, and an assessment of your overall dental status.

Risks involved in participating in this study
There are no risks involved in this study.

Benefits of being involved in the study
There are no direct benefits of taking part in this study

Will participation in this study affect my treatment?
Participating will in no way affect the treatment or services provided to you.

What happens if I want to withdraw from this study?
You are perfectly free to withdraw from this research project at any time you want to – even in the middle of completing a questionnaire. Such withdrawal will in no way affect the treatment or services provided to you at this clinic.

Confidentiality

We assure you that all information gathered during the course of this research project will be kept completely confidential. Only Dr. Allison (the principal researcher involved in the project) plus the research assistant gathering the data will have access to the data, which will be kept locked in Dr. Allison's office. All data will be identified through a code number so we will not know to whom the data relates. The results of the research will be published in scientific medical journals in an anonymous form.

Further information

If you would like any more information or have any questions related to this study, please do not hesitate to call the principal investigator for this project Dr. Paul Allison (514-398-7203 ext. 00045). In addition, if you have any questions concerning your rights as a research subject, you may contact the Montreal Children's Hospital ombudsman Elisabeth Gibbon (514-412-4400 ext. 22223).

Consent

I have read and had explained to me the information concerning this study and agree to participate. It has been indicated to me that participation is entirely voluntary and that I may withdraw at any moment without it in any way affecting my treatment.

Name ....

Signature.. Date................................

Witness name.....................................

Signature.. Date................................

10.2. English consent form (private clinic) for adult subjects

DEVELOPMENT OF A QUESTIONNAIRE TO ASSESS ORAL HEALTH PROBLEMS IN PEOPLE WITH DOWN SYNDROME

Dr. P.J. Allison
Faculty of Dentistry, McGill University

CONSENT FORM

Introduction

We are interested in finding out about the oral and dental health problems experienced by people with Down syndrome in Canada. There are currently no data concerning the dental health and the dental care provision for people with Down syndrome in this country. However, evidence from other countries suggests that people with Down syndrome have a high level of oral and dental disease but have major problems accessing dental services. In order to assess the extent of oral problems, this study requires that we seek those individuals with and without Down syndrome in order to compare the two groups.

If you agree to participate, what will that involve?

PARTICIPATION IS ENTIRELY VOLUNTARY. If you agree to participate, all that would be required is for you to complete our questionnaire with respect to yourself and allow the clinic dentist (Dr. Bonin) to examine you, and collect data for the study that he often routinely collects anyway. Such data would include the number of fillings you may have, and an assessment of your overall dental status.

Risks involved in participating in this study

There are no risks involved in this study.

Benefits of being involved in the study

There are no direct benefits of taking part in this study

Will participation in this study affect my treatment?

Participating will in no way affect the treatment or services provided to you.

What happens if I want to withdraw from this study?

You are perfectly free to withdraw from this research project at any time you want to – even in the middle of completing a questionnaire. Such withdrawal will in no way affect the treatment or services provided to you at this clinic.

Confidentiality

We assure you that all information gathered during the course of this research project will be kept completely confidential. Only Dr. Allison (the principal researcher involved in the project) plus the research assistant gathering the data will have access to the data, which will be kept locked in Dr. Allison's office. All data will be identified through a code number so we will not know to whom the data relates. The results of the research will be published in scientific medical journals in an anonymous form.

Further information

If you would like any more information or have any questions related to this study, please do not hesitate to call the principal investigator for this project Dr. Paul Allison (514 398 7203 ext. 00045).

Consent

I have read and had explained to me the information concerning this study and agree to participate. It has been indicated to me that participation is entirely voluntary and that I may withdraw at any moment without it in any way affecting my treatment.

Name ....

Signature.. Date.................................

Witness name.....................................

Signature.. Date.................................

10.3. French consent form (MCH) for adult subjects

ÉLABORATION D'UN QUESTIONNAIRE DANS LE BUT D'ÉVALUER LES PROBLÈMES DE SANTE BUCCALE CHEZ LES INDIVIDUS TRISOMIQUES (SYNDROME DE DOWN)

Dr P.J. Allison
Faculté de médecine dentaire, Université McGill

FORMULAIRE DE CONSENTEMENT

Introduction

Nous cherchons à connaître l'état de santé buccale et dentaire des individus trisomiques au Canada. Il n'existe actuellement aucune documentation sur la santé dentaire et le niveau des soins dentaires desservant cette particulière population canadienne. Pourtant, d'après la documentation existant dans d'autres pays, il semblerait que les personnes avec le syndrome de Down auraient des problèmes buccaux et dentaires importants, et que de plus, elles éprouveraient de grandes difficultés à accéder aux soins dentaires dont elles auraient besoin. Alors pour mesurer la mesure de ces problèmes buccaux et dentaires, il faut que nous trouvions des individus sans et avec le syndrome de Down. Cette méthode va nous aider à comparer les deux groupes.

Si vous acceptez de participer, quelles en seront les conséquences?

VOTRE PARTICIPATION EST ENTIEREMENT VOLONTAIRE. Si vous acceptez de participer à ce projet, tout ce que vous aurez à faire sera de répondre à notre questionnaire-essai concernant vous-même et à permettre au dentiste de la clinique (Dr. Schwartz) de faire un examen dentaire et de rassembler les résultats pour notre enquête. Ce sont des examens de routine, où le nombre des dents cariées et restaurées sont listées ainsi que la santé des gencives et la présence ou non de plaque dentaire.

Risques inhérents à la participation à cette enquête

Il n'y a aucun risque associé à cette enquête

Bénéfices à retirer de la participation à cette enquête

Il n'y a aucun bénéfice direct à retirer pour vous de la participation à cette enquête

Ma participation à cette enquête pourra-t-elle affecter les traitements dentaires que je reçois ailleurs?

Votre participation n'affectera en rien vos traitements

Qu'arrivera-t-il si je veux me retirer de l'enquête?
Vous êtes parfaitement libre de vous retirer de ce projet de recherche à n'importe quel moment. Votre retrait n'affectera en rien vos traitements ou tout autre service qui vous est offert dans votre clinique.

Confidentialité
Nous pouvons vous assurer que toutes les informations réunies au cours de ce projet de recherche seront gardées en stricte confidentialité. Dr Allison (le chercheur principal responsable du projet) ainsi que l'assistance de recherche qui collecte les données auront accès à ces renseignements qui seront, eux-mêmes, gardés sous clefs dans son bureau. Chaque dossier ne sera identifiable que par un numéro codé, et nous ne saurons pas à qui ces dossiers appartiennent. Les résultats de l'étude qui seront publiés dans des journaux médicaux scientifiques seront sous une forme complètement anonyme.

Autre information
Si vous cherchez la réponse à d'autres questions que vous pourriez vous poser, n'hésitez pas à appeler le Dr. Paul Allison (514-398-7203 ext. 00045).

Consentement éclairé
J'ai bien lu ce formulaire de consentement et on m'a également donné toutes les informations nécessaires à cette enquête, et j'accepte d'y participer. Je comprends que cette participation est tout à fait volontaire et que je peux l'interrompre à n'importe quel moment sans qu'il n'en résulte de préjudice pour mes traitements.

Nom ....

Signature .... Date ....

Nom du témoin ....

Signature .... Date ....

10.4. French consent form (private clinic) for adult subjects

ÉLABORATION D'UN QUESTIONNAIRE DANS LE BUT D'ÉVALUER LES PROBLÈMES DE SANTE BUCCALE CHEZ LES INDIVIDUS TRISOMIQUES (SYNDROME DE DOWN)

Dr. P.J. Allison

Faculté de médecine dentaire, Université McGill

FORMULAIRE DE CONSENTEMENT

Introduction

Nous cherchons à connaître l'état de santé buccale et dentaire des individus trisomiques au Canada. Il n'existe actuellement aucune documentation sur la santé dentaire et le niveau des soins dentaires desservant cette particulière population canadienne. Pourtant, d'après la documentation existant dans d'autres pays, il semblerait que les personnes avec le syndrome de Down auraient des problèmes buccaux et dentaires importants, et que de plus, elles éprouveraient de grandes difficultés à accéder aux soins dentaires dont elles auraient besoin. Alors pour mesurer la mesure de ces problèmes buccaux et dentaires, il faut que nous trouvions des individus sans et avec le syndrome de Down. Cette méthode va nous aider à comparer les deux groupes.

Si vous acceptez de participer, quelles en seront les conséquences?

VOTRE PARTICIPATION EST ENTIEREMENT VOLONTAIRE. Si vous acceptez de participer à ce projet, tout ce que vous aurez à faire sera de répondre à notre questionnaire-essai concernant vous-même et à permettre au dentiste de la clinique (Dr. Bonin) de faire un examen dentaire et de rassembler les résultats pour notre enquête. Ce sont des examens de routine, où le nombre des dents cariées et restaurées sont listées ainsi que la santé des gencives et la présence ou non de plaque dentaire.

Risques inhérents à la participation à cette enquête

Il n'y a aucun risque associé à cette enquête

Bénéfices à retirer de la participation à cette enquête

Il n'y a aucun bénéfice direct à retirer pour vous de la participation à cette enquête

Ma participation à cette enquête pourra-t-elle affecter les traitements dentaires que je reçois ailleurs?

Votre participation n'affectera en rien vos traitements

Qu'arrivera-t-il si je veux me retirer de l'enquête?
Vous êtes parfaitement libre de vous retirer de ce projet de recherche à n'importe quel moment. Votre retrait n'affectera en rien vos traitements ou tout autre service qui vous est offert dans votre clinique.

Confidentialité
Nous pouvons vous assurer que toutes les informations réunies au cours de ce projet de recherche seront gardées en stricte confidentialité. Dr Allison (le chercheur principal responsable du projet) ainsi que l'assistance de recherche qui collecte les données auront accès à ces renseignements qui seront, eux-mêmes, gardés sous clefs dans son bureau. Chaque dossier ne sera identifiable que par un numéro codé, et nous ne saurons pas à qui ces dossiers appartiennent. Les résultats de l'étude qui seront publiés dans des journaux médicaux scientifiques seront sous une forme complètement anonyme.

Autre information
Si vous cherchez la réponse à d'autres questions que vous pourriez vous poser, n'hésitez pas à appeler le Dr. Paul Allison (514-398-7203 ext. 00045).

Consentement éclairé
J'ai bien lu ce formulaire de consentement et on m'a également donné toutes les informations nécessaires à cette enquête, et j'accepte d'y participer. Je comprends que cette participation est tout à fait volontaire et que je peux l'interrompre à n'importe quel moment sans qu'il n'en résulte de préjudice pour mes traitements.

Nom ....

Signature .... Date ....

Nom du témoin ....

Signature .... Date ....

10.5. English consent form (MCH) for parents

DEVELOPMENT OF A QUESTIONNAIRE TO ASSESS ORAL HEALTH PROBLEMS IN PEOPLE WITH DOWN SYNDROME

Dr. P.J. Allison and Dr. S. Schwartz
Faculty of Dentistry, McGill University

CONSENT FORM

Introduction

We are interested in finding out about the oral and dental health problems experienced by people with Down syndrome in Canada. There are currently no data concerning the dental health and the dental care provision for people with Down syndrome in this country. However, evidence from other countries suggests that people with Down syndrome have a high level of oral and dental disease but have major problems accessing dental services. We are therefore developing a questionnaire designed to be completed by the parents/guardians of people with Down syndrome and we need your help to complete the task. This study requires that we seek those individuals with and without Down syndrome in order to compare the two groups.

If you agree to participate, what will that involve?

PARTICIPATION IS ENTIRELY VOLUNTARY. If you agree to participate, all that would be required is for you to complete our test questionnaire with respect to your child and allow the clinic dentist (Dr. Schwartz) to examine your child collecting data for the study that she often routinely collects anyway. Such data would include the number of fillings your child has, whether or not his/her gums bleed and whether or not there is any plaque on his/her teeth.

Risks involved in participating in this study

There are no risks involved in this study.

Benefits of being involved in the study

There are no direct benefits of taking part in this study

Will participation in this study affect my treatment?

Participating will in no way affect the treatment or services provided to your child.

What happens if I want to withdraw from this study?

You are perfectly free to withdraw from this research project at any time you want to – even in the middle of completing a questionnaire. Such withdrawal will in no way affect the treatment or services provided to your child at this clinic.

Confidentiality

We assure you that all information gathered during the course of this research project will be kept completely confidential. Only Dr. Allison (the principal researcher involved in the project) and the research assistant gathering the data will have access to the data, which will be kept locked in Dr. Allison's office. All data will be identified through a code number so we will not know to whom the data relates. The results of the research will be published in scientific medical journals in an anonymous form.

Further information

If you would like any more information or have any questions related to this study, please do not hesitate to call the principal investigator for this project Dr. Paul Allison (514 398 7203 ext. 00045). In addition, if you have any questions concerning your rights as a research subject, you may contact the Montreal Children's Hospital ombudsman Elisabeth Gibbon (514 412 4400 ext. 22223).

Consent

I have read and had explained to me the information concerning this study and agree to participate. It has been indicated to me that participation is entirely voluntary and that I may withdraw at any moment without it in any way affecting my child's treatment.

Name ....

Signature... Date.................................

Witness name......................................

Signature... Date.................................

10.6. English consent form (private clinic) for parents

DEVELOPMENT OF A QUESTIONNAIRE TO ASSESS ORAL HEALTH PROBLEMS IN PEOPLE WITH DOWN SYNDROME

Dr. P.J. Allison
Faculty of Dentistry, McGill University

CONSENT FORM

Introduction
We are interested in finding out about the oral and dental health problems experienced by people with Down syndrome in Canada. There are currently no data concerning the dental health and the dental care provision for people with Down syndrome in this country. However, evidence from other countries suggests that people with Down syndrome have a high level of oral and dental disease but have major problems accessing dental services. In order to assess the extent of oral problems, this study requires that we seek those individuals with and without Down syndrome in order to compare the two groups.

If you agree to participate, what will that involve?
PARTICIPATION IS ENTIRELY VOLUNTARY. If you agree to participate, all that would be required is for you to complete our questionnaire with respect to your child and allow the clinic dentist (Dr. Bonin) to examine him/her and collect data for the study that he often routinely collects anyway. Such data would include the number of fillings, and an assessment of your child's overall dental status.

Risks involved in participating in this study
There are no risks involved in this study.

Benefits of being involved in the study
There are no direct benefits of taking part in this study

Will participation in this study affect my treatment?
Participating will in no way affect the treatment or services provided to you.

What happens if I want to withdraw from this study?
You are perfectly free to withdraw from this research project at any time you want to – even in the middle of completing a questionnaire. Such withdrawal will in no way affect the treatment or services provided to your child at this clinic.

Confidentiality

We assure you that all information gathered during the course of this research project will be kept completely confidential. Only Dr. Allison (the principal researcher involved in the project) plus the research assistant gathering the data will have access to the data, which will be kept locked in Dr. Allison's office. All data will be identified through a code number so we will not know to whom the data relates. The results of the research will be published in scientific medical journals in an anonymous form.

Further information

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Consent

I have read and had explained to me the information concerning this study and agree to participate. It has been indicated to me that participation is entirely voluntary and that I may withdraw at any moment without it in any way affecting my child's treatment.

Name ....

Signature.. Date.................................

Witness name.....................................

Signature.. Date.................................

10.7. French consent form (MCH) for parents

ÉLABORATION D'UN QUESTIONNAIRE DANS LE BUT D'ÉVALUER LES PROBLÈMES DE SANTE BUCCALE CHEZ LES INDIVIDUS TRISOMIQUES (SYNDROME DE DOWN)

Dr P.J. Allison et Dr S. Schwartz
Faculté de médecine dentaire, Université McGill

FORMULAIRE DE CONSENTEMENT

Introduction

Nous cherchons à connaître l'état de santé buccale et dentaire des individus trisomiques au Canada. Il n'existe actuellement aucune documentation sur la santé dentaire et le niveau des soins dentaires desservant cette particulière population canadienne. Pourtant, d'après la documentation existant dans d'autres pays, il semblerait que les personnes avec le syndrome de Down auraient des problèmes buccaux et dentaires importants, et que de plus, elles éprouveraient de grandes difficultés à accéder aux soins dentaires dont elles auraient besoin. Alors pour mesurer la mesure de ces problèmes buccaux et dentaires, il faut que nous trouvions des individus sans et avec le syndrome de Down. Cette méthode va nous aider à comparer les deux groupes.

Si vous acceptez de participer, quelles en seront les conséquences?

VOTRE PARTICIPATION EST ENTIEREMENT VOLONTAIRE. Si vous acceptez de participer à ce projet, tout ce que vous aurez à faire sera de répondre à notre questionnaire-essai concernant votre enfant et à permettre au dentiste de la clinique (Dr. Schwartz) de faire un examen dentaire et de rassembler les résultats pour notre enquête. Ce sont des examens de routine, où le nombre des dents cariées et restaurées sont listées ainsi que la santé des gencives et la présence ou non de plaque dentaire.

Risques inhérents à la participation à cette enquête

Il n'y a aucun risque associé à cette enquête

Bénéfices à retirer de la participation à cette enquête

Il n'y a aucun bénéfice direct à retirer pour vous de la participation à cette enquête

Ma participation à cette enquête pourra-t-elle affecter les traitements dentaires que je reçois ailleurs?

Votre participation n'affectera en rien vos traitements

Qu'arrivera-t-il si je veux me retirer de l'enquête?
Vous êtes parfaitement libre de vous retirer de ce projet de recherche à n'importe quel moment. Votre retrait n'affectera en rien vos traitements ou tout autre service qui vous est offert dans votre clinique

Confidentialité
Nous pouvons vous assurer que toutes les informations réunies au cours de ce projet de recherche seront gardées en stricte confidentialité. Dr Allison (le chercheur principal responsable du projet) ainsi que l'assistance de recherche qui collecte les données auront accès à ces renseignements qui seront, eux-mêmes, gardés sous clefs dans son bureau. Chaque dossier ne sera identifiable que par un numéro codé, et nous ne saurons pas à qui ces dossiers appartiennent. Les résultats de l'étude qui seront publiés dans des journaux médicaux scientifiques seront sous une forme complètement anonyme.

Autre information
Si vous cherchez la réponse à d'autres questions que vous pourriez vous poser, n'hésitez pas à appeler le Dr. Paul Allison (514 398 7203 ext 00045).

Consentement éclairé
J'ai bien lu ce formulaire de consentement et on m'a également donné toutes les informations nécessaires reliées à cette enquête, et j'accepte d'y participer. Je comprends que cette participation est tout à fait volontaire et que je peux l'interrompre à n'importe quel moment sans qu'il n'en résulte de préjudice pour les traitements de mon enfant.

Nom ....

Signature .... Date ....

Nom du témoin ....

Signature .... Date ....

10.8. French consent form (private clinic) for parents

ÉLABORATION D'UN QUESTIONNAIRE DANS LE BUT D'ÉVALUER LES PROBLÈMES DE SANTE BUCCALE CHEZ LES INDIVIDUS TRISOMIQUES (SYNDROME DE DOWN)

Dr. P.J. Allison

Faculté de médecine dentaire, Université McGill

FORMULAIRE DE CONSENTEMENT

Introduction
Nous cherchons à connaître l'état de santé buccale et dentaire des individus trisomiques au Canada. Il n'existe actuellement aucune documentation sur la santé dentaire et le niveau des soins dentaires desservant cette particulière population canadienne. Pourtant, d'après la documentation existant dans d'autres pays, il semblerait que les personnes avec le syndrome de Down auraient des problèmes buccaux et dentaires importants, et que de plus, elles éprouveraient de grandes difficultés à accéder aux soins dentaires dont elles auraient besoin. Alors pour mesurer la mesure de ces problèmes buccaux et dentaires, il faut que nous trouvions des individus sans et avec le syndrome de Down. Cette méthode va nous aider à comparer les deux groupes.
Si vous acceptez de participer, quelles en seront les conséquences?
VOTRE PARTICIPATION EST ENTIEREMENT VOLONTAIRE. Si vous acceptez de participer à ce projet, tout ce que vous aurez à faire sera de répondre à notre questionnaire-essai concernant votre enfant et à permettre au dentiste de la clinique (Dr. Bonin) de faire un examen dentaire et de rassembler les résultats pour notre enquête. Ce sont des examens de routine, où le nombre des dents cariées et restaurées sont listées ainsi que la santé des gencives et la présence ou non de plaque dentaire.

Risques inhérents à la participation à cette enquête
Il n'y a aucun risque associé à cette enquête

Bénéfices à retirer de la participation à cette enquête
Il n'y a aucun bénéfice direct à retirer pour vous de la participation à cette enquête

Ma participation à cette enquête pourra-t-elle affecter les traitements dentaires que je reçois ailleurs?
Votre participation n'affectera en rien vos traitements

Qu'arrivera-t-il si je veux me retirer de l'enquête?
Vous êtes parfaitement libre de vous retirer de ce projet de recherche à n'importe quel moment. Votre retrait n'affectera en rien vos traitements ou tout autre service qui vous est offert dans votre clinique

Confidentialité

Nous pouvons vous assurer que toutes les informations réunies au cours de ce projet de recherche seront gardées en stricte confidentialité. Dr Allison (le chercheur principal responsable du projet) ainsi que l'assistance de recherche qui collecte les données auront accès à ces renseignements qui seront, eux-mêmes, gardés sous clefs dans son bureau. Chaque dossier ne sera identifiable que par un numéro codé, et nous ne saurons pas à qui ces dossiers appartiennent. Les résultats de l'étude qui seront publiés dans des journaux médicaux scientifiques seront sous une forme complètement anonyme.

Autre information

Si vous cherchez la réponse à d'autres questions que vous pourriez vous poser, n'hésitez pas à appeler le Dr. Paul Allison (514 398 7203 ext 00045).

Consentement éclairé

J'ai bien lu ce formulaire de consentement et on m'a également donné toutes les informations nécessaires reliées à cette enquête, et j'accepte d'y participer. Je comprends que cette participation est tout à fait volontaire et que je peux l'interrompre à n'importe quel moment sans qu'il n'en résulte de préjudice pour les traitements de mon enfant.

Nom ....

Signature .... Date ....

Nom du témoin ....

Signature .... Date ....

10.9. Dental data worksheet

McGill University Down Syndrome Research Study
Dental data sheet

Subject ID ______________________

Date of questionnaire completion _______________ (dd/mm/yyyy)

---

1. **Number of teeth present** ___________
2. **Number of decayed teeth (D)** ___________
3. **Number of missing teeth (M)** ___________
    - **Number of teeth missing congenitally** ___________
    - **Number of teeth missing because of acquired disease** ___________
    - **Number of teeth missing – reason unknown** ___________
4. **Number of filled teeth (F)** ___________
5. **Total DMFT** ___________
6. **Gingival index (GI)**

Tooth	GI
Buccal surface last upper occluding molar on left	_____
Lingual surface last lower occluding molar on left	_____
Buccal surface of upper left central incisor	_____
Buccal surface of lower right central incisor	_____

0 = normal gingiva

1 = mild inflam'n (slight colour change, no eodema, no bleeding on probing)

2 = moderate inflam'n (red, oedematous, glazing and bleeding on probing)

3 = severe inflam'n (marked redness and oedema, ulceration, tendency to spontaneous bleeding)

7. **Plaque index (PI)**

Tooth	PI
Buccal surface last upper occluding molar on left	_____
Lingual surface last lower occluding molar on left	_____
Buccal surface of upper left central incisor	_____
Buccal surface of lower right central incisor	_____

0 = no plaque

1 = plaque on gingival third of tooth only

2 = plaque on gingival and mid third of tooth

3 = plaque on gingival, mid and coronal third of tooth

10.10. Caries diagnosis criteria

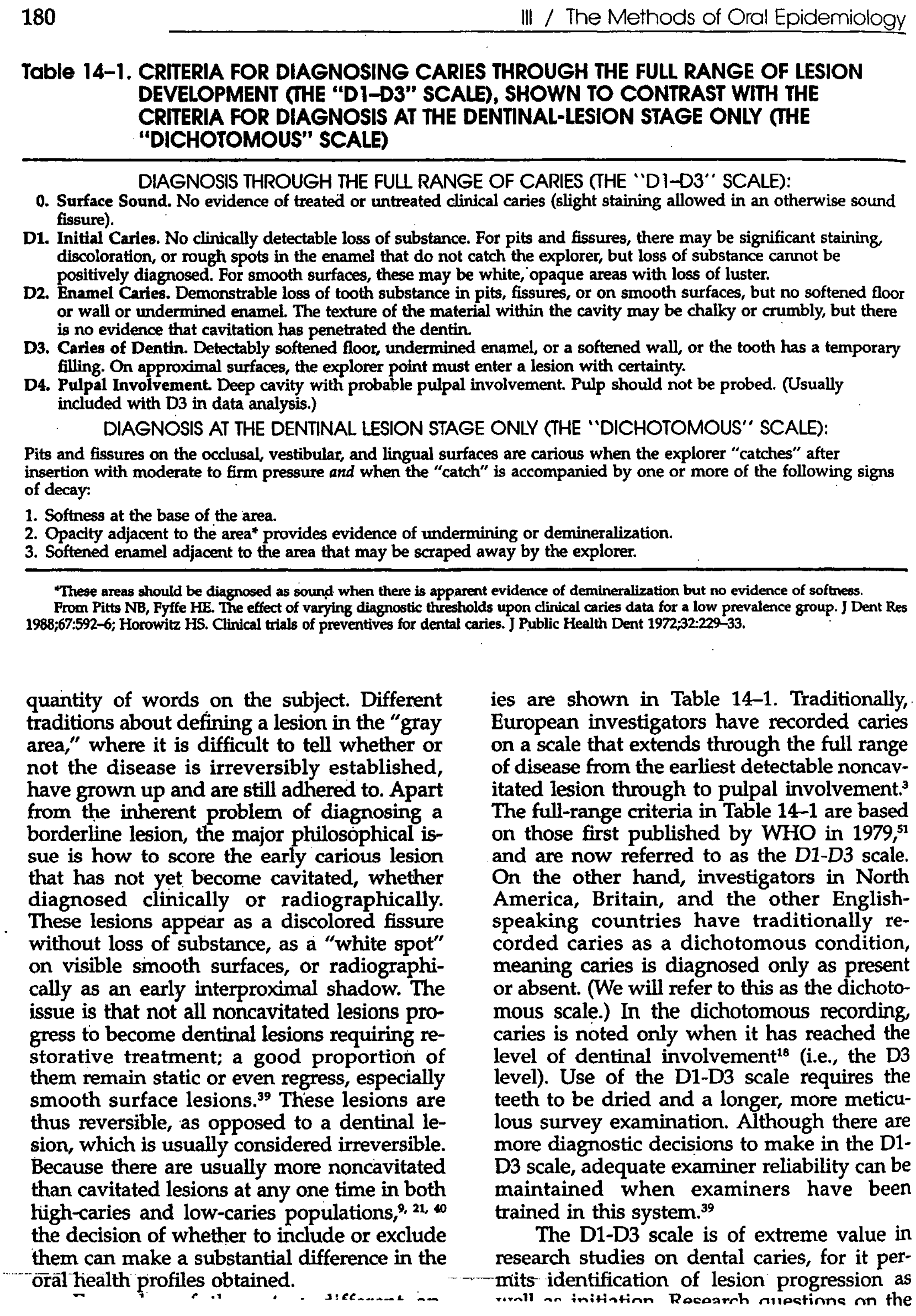

Table 14-1. CRITERIA FOR DIAGNOSING CARIES THROUGH THE FULL RANGE OF LESION DEVELOPMENT (THE "D1–D3" SCALE), SHOWN TO CONTRAST WITH THE CRITERIA FOR DIAGNOSIS AT THE DENTINAL-LESION STAGE ONLY (THE "DICHOTOMOUS" SCALE)

DIAGNOSIS THROUGH THE FULL RANGE OF CARIES (THE "D1–D3" SCALE):

0. **Surface Sound.** No evidence of treated or untreated clinical caries (slight staining allowed in an otherwise sound fissure).

D1. **Initial Caries.** No clinically detectable loss of substance. For pits and fissures, there may be significant staining, discoloration, or rough spots in the enamel that do not catch the explorer, but loss of substance cannot be positively diagnosed. For smooth surfaces, these may be white, opaque areas with loss of luster.

D2. **Enamel Caries.** Demonstrable loss of tooth substance in pits, fissures, or on smooth surfaces, but no softened floor or wall or undermined enamel. The texture of the material within the cavity may be chalky or crumbly, but there is no evidence that cavitation has penetrated the dentin.

D3. **Caries of Dentin.** Detectably softened floor, undermined enamel, or a softened wall, or the tooth has a temporary filling. On approximal surfaces, the explorer point must enter a lesion with certainty.

D4. **Pulpal Involvement.** Deep cavity with probable pulpal involvement. Pulp should not be probed. (Usually included with D3 in data analysis.)

DIAGNOSIS AT THE DENTINAL LESION STAGE ONLY (THE "DICHOTOMOUS" SCALE):

Pits and fissures on the occlusal, vestibular, and lingual surfaces are carious when the explorer "catches" after insertion with moderate to firm pressure *and* when the "catch" is accompanied by one or more of the following signs of decay:

1. Softness at the base of the area.
2. Opacity adjacent to the area* provides evidence of undermining or demineralization.
3. Softened enamel adjacent to the area that may be scraped away by the explorer.

*These areas should be diagnosed as sound when there is apparent evidence of demineralization but no evidence of softness.

From Pitts NB, Fyffe HE. The effect of varying diagnostic thresholds upon clinical caries data for a low prevalence group. J Dent Res 1988;67:592–6; Horowitz HS. Clinical trials of preventives for dental caries. J Public Health Dent 1972;32:229–33.

quantity of words on the subject. Different traditions about defining a lesion in the "gray area," where it is difficult to tell whether or not the disease is irreversibly established, have grown up and are still adhered to. Apart from the inherent problem of diagnosing a borderline lesion, the major philosophical issue is how to score the early carious lesion that has not yet become cavitated, whether diagnosed clinically or radiographically. These lesions appear as a discolored fissure without loss of substance, as a "white spot" on visible smooth surfaces, or radiographically as an early interproximal shadow. The issue is that not all noncavitated lesions progress to become dentinal lesions requiring restorative treatment; a good proportion of them remain static or even regress, especially smooth surface lesions.[39] These lesions are thus reversible, as opposed to a dentinal lesion, which is usually considered irreversible. Because there are usually more noncavitated than cavitated lesions at any one time in both high-caries and low-caries populations,[9, 21, 40] the decision of whether to include or exclude them can make a substantial difference in the oral health profiles obtained.

ies are shown in Table 14–1. Traditionally, European investigators have recorded caries on a scale that extends through the full range of disease from the earliest detectable noncavitated lesion through to pulpal involvement.[3] The full-range criteria in Table 14–1 are based on those first published by WHO in 1979,[51] and are now referred to as the *D1-D3* scale. On the other hand, investigators in North America, Britain, and the other English-speaking countries have traditionally recorded caries as a dichotomous condition, meaning caries is diagnosed only as present or absent. (We will refer to this as the dichotomous scale.) In the dichotomous recording, caries is noted only when it has reached the level of dentinal involvement[18] (i.e., the D3 level). Use of the D1-D3 scale requires the teeth to be dried and a longer, more meticulous survey examination. Although there are more diagnostic decisions to make in the D1-D3 scale, adequate examiner reliability can be maintained when examiners have been trained in this system.[39]

The D1-D3 scale is of extreme value in research studies on dental caries, for it permits identification of lesion progression as well as initiation. Research questions on the

10.11. English Oral Assessment in Down syndrome (OADS) questionnaire

McGill UNIVERSITY / CANADIAN DOWN SYNDROME SOCIETY RESEARCH PROJECT

Please complete the following questionnaire with respect to your child with Down syndrome by putting a tick or cross in the box corresponding with your response to each question. **When we say "your child" in the question, we are always referring to your child with Down syndrome.** May we remind you that all the information you give is **completely anonymous** and will be kept **strictly confidential.**

1. Are you your child's ....
   ☐ Mother ☐ Father ☐ Brother/sister ☐ Other guardian

2. At what level did you finish your formal education?
   ☐ School/high school ☐ college ☐ university

3. In which village/town/city and province/territory do you live?
   Village/Town/City: ______________________
   Province/Territory: ______________________

4. Would you describe this location as ....
   ☐ urban ☐ suburban ☐ rural

5. Where does your child live?
   ☐ with you
   ☐ independently
   ☐ in some form of communal accommodation
   ☐ other or combination (please specify ______________________)

6. If your child does not live with you, where does he/she live?
   Village/Town/City: ______________________
   Province/Territory: ______________________

7. Which of the following best describes your child's ethnic background?
   ☐ white
   ☐ black
   ☐ native North American
   ☐ Asian
   ☐ other (please specify ______________________)

8. How old is your child? _________

9. Is your child: ☐ male ☐ female

10. Does you child have any brothers or sisters? ☐ yes ☐ no

11. Which of the following best describes your child's daily activities?

☐ he/she stays principally at home
☐ he/she attends a mainstream school
☐ he/she attends some form of specialized school
☐ he/she attends a day centre
☐ he/she works in a "protected environment"
☐ he/she works in the community
☐ other or combination (please specify ________________________)

12. If your child suffers from any of the medical problems listed below, please note the frequency with which he/she sees a doctor or a specialist concerning this problem.

	At least once a month	Several times a year	Once a year	Less than once a year	**Not relevant**
▪ Heart problems	☐	☐	☐	☐	☐
▪ Immunological problems	☐	☐	☐	☐	☐
▪ Ear infections, tonsilitis, etc..	☐	☐	☐	☐	☐
▪ Speech problems	☐	☐	☐	☐	☐
▪ Visual problems	☐	☐	☐	☐	☐
▪ Orofacial or dental problems	☐	☐	☐	☐	☐

	Yes	No	I don't know
13. Does your child's medical care take a lot of time compared to your other obligations?	☐	☐	☐
14. Does your child attend a <u>medical</u> clinic specialized in the care of people with Down syndrome?	☐	☐	☐
15. Does your child attend a <u>dental</u> clinic specialized in the care of people with Down syndrome?	☐	☐	☐
16. Is it difficult to find a <u>doctor</u> to care for your child?	☐	☐	☐
17. Is it difficult to find a <u>dentist</u> to care for your child?	☐	☐	☐
18. Do you visit a dentist yearly with your child?	☐	☐	☐

19. Roughly, how far is it to the dentist you normally visit with your child? ______ km

20. Do you (or did you) ever regularly visit an orthodontist with your child?

☐ Yes ☐ No

If yes, approximately how far do/did you have to travel? __________ km

		Not at all	A little	Quite a lot	Very much
21.	Does your child find it difficult to bite into a whole, unpeeled apple?	☐	☐	☐	☐
22.	Does your child have difficulty chewing sliced pieces of apple?	☐	☐	☐	☐
23.	Does your child have difficulty chewing pieces of meat?	☐	☐	☐	☐
24.	During a meal, does your child tend to bring up wind (belch or burp)?	☐	☐	☐	☐
25.	Normally, is your child constipated?	☐	☐	☐	☐
26.	During the day, does your child often have his/her tongue protruding from his/her mouth?	☐	☐	☐	☐
27.	Do your childs' gums bleed easily?	☐	☐	☐	☐
28.	Does your child have bad breath?	☐	☐	☐	☐
29.	Does your child grind his/her teeth?	☐	☐	☐	☐
30.	During chewing, does food spill out of your child's mouth?	☐	☐	☐	☐
31.	Does your child have a strong gag reflex (i.e. he/she retches or is sick easily if something is placed in his/her mouth)?	☐	☐	☐	☐
32.	Does your child sleep with his/her mouth open?	☐	☐	☐	☐

		Yes	No	I don't know
33.	When your child was breast or bottle fed, did he/she have difficulty suckling?	☐	☐	☐
34.	Did your child have difficulty changing from pureed food to spoon-fed solids?	☐	☐	☐

35.	Did your child's milk teeth take a long time to fall out?	☐	☐	☐
36.	Has your child ever undergone dental treatment?	☐	☐	☐
37.	Has your child ever taken fluoride tablets or drops?	☐	☐	☐
38.	Has your child ever received any form of fluoride treatment at the dentist?	☐	☐	☐
		Yes	No	I don't know
39.	Has your child ever had a tooth extracted?	☐	☐	☐
40.	Has your child ever had a filling?	☐	☐	☐
41.	Has your child ever been put to sleep (with a general anaesthetic) for a dental procedure?	☐	☐	☐
42.	Does your child have, or has he/she ever had, a denture, a bridge, or a crown?	☐	☐	☐
43.	Is your child undergoing, or has he/she ever undergone, orthodontic treatment (treatment to correct tooth position)?	☐	☐	☐
44.	Are your child's teeth brushed daily (by him/her or by somebody else)?	☐	☐	☐
45.	Is your child able to eat all types of foods?	☐	☐	☐
46.	Is your child capable of cleaning his/her teeth his/herself?	☐	☐	☐
47.	Is it difficult to judge whether your child is in pain?	☐	☐	☐
48.	Is it difficult to judge where you child has pain?	☐	☐	☐
49.	In as much as you can judge, has your child ever suffered from painful dental problems?	☐	☐	☐
50.	Is your child able to talk?	☐	☐	☐
51.	Is your child understood by people outside the immediate family when he/she talks?	☐	☐	☐
52.	Is your child able to pronounce the syllable "t" (as in "toy")?	☐	☐	☐

		Yes	No	I don't know
53.	Is your child able to pronounce the syllable "g" (as in "grandma")?	☐	☐	☐

		Very poor	Poor	Reasonable	Good	Excellent
54.	How would you describe the general health of your child?	☐	☐	☐	☐	☐
55.	How would you describe the oral or dental health of your child?	☐	☐	☐	☐	☐

10.12. French Oral Assessment in Down syndrome (OADS) questionnaire

UNIVERSITÉ McGILL / SOCIÉTÉ CANADIENNE DU SYNDROME DE DOWN
PROJET DE RECHERCHE

S'il-vous-plaît, veuillez compléter le questionnaire suivant pour votre enfant trisomique. Pour chaque question, mettez une croix ou un crochet dans la case correspondant à votre réponse. **Chaque fois que vous voyez le terme "votre enfant" il s'agit de votre enfant trisomique**. Nous aimerions vous rappeler que toutes les informations que nous recueillons grâce à ce questionnaire demeureront **anonymes** et **strictement confidentielles.**

1. Quel est votre lien de parenté avec l'enfant ?
☐ mère ☐ père ☐ frère/sœur ☐ autre tuteur

2. Quel est votre niveau de scolarité ?
☐ primaire ou secondaire ☐ cégep/collège ☐ université

3. Dans quel village ou ville et quelle province ou territoire vivez-vous ?
village/ville : ______________________________
province/territoire : ________________________

4. Où cet endroit est-il situé ?
☐ en ville ☐ en banlieue ☐ à la campagne

9. Où vit votre enfant ?
☐ avec vous
☐ seul
☐ dans un logement communautaire
☐ autre ou une combinaison des choix qui précèdent (spécifiez s.v.p.____________)

10. Si votre enfant ne vit pas avec vous, où vit-il ?
village/ville : _________________________
province/territoire : ________________________

11. Lequel des choix suivants décrits le mieux l'origine ethnique de votre enfant ?
☐ blanc
☐ noir
☐ amérindien
☐ asiatique
☐ autre (spécifiez s.v.p. _________________________________)

12. Quel âge a votre enfant ? __________

9. Quel est le sexe de votre enfant ? ☐ masculin ☐ féminin

10. Votre enfant a-t-il des frères ou des sœurs ? ☐ oui ☐ non

11. Qu'est-ce qui décrit le mieux le quotidien de votre enfant ?
☐ il reste principalement à la maison
☐ il fréquente une école régulière
☐ il fréquente une école spécialisée
☐ il fréquente un centre de jour
☐ il travaille dans un environnement protégé
☐ il travaille dans la communauté
☐ autre ou une combinaison des choix qui précèdent (spécifiez s.v.p.______________)

12. Si votre enfant souffre d'un ou plusieurs problèmes mentionnés dans la liste suivante, s.v.p. indiquez le nombre de fois que ces problèmes l'obligent à consulter un médecin ou un spécialiste.

	Au moins une fois par mois	Plusieurs fois par année	Une fois par année	Moins d'une fois par année	Ne s'applique pas
▪ problèmes cardiaques	☐	☐	☐	☐	☐
▪ problèmes immunitaires (ex: leucémie)	☐	☐	☐	☐	☐
▪ infections aux oreilles, amygdalite etc.	☐	☐	☐	☐	☐
▪ problèmes de langage	☐	☐	☐	☐	☐
▪ problèmes visuals	☐	☐	☐	☐	☐
▪ problèmes bucco-dentaires	☐	☐	☐	☐	☐

		Oui	Non	Je ne sais pas
13.	Est-ce que les soins médicaux apportés à votre enfant prennent beaucoup de temps comparé à vos autres obligations ?	☐	☐	☐
14.	Est-ce que votre enfant fréquente une clinique <u>médicale</u> spécialisée dans les soins apportés aux personnes trisomiques ?	☐	☐	☐
15.	Est-ce que votre enfant fréquente une clinique <u>dentaire</u> spécialisée dans les soins apportés aux personnes trisomiques ?	☐	☐	☐
16.	Est-ce difficile de trouver un médecin pour prendre soin de votre enfant ?	☐	☐	☐
17.	Est-ce difficile de trouver un dentiste pour prendre soin de votre enfant ?	☐	☐	☐
18.	Allez-vous chez le dentiste chaque année avec votre enfant ?	☐	☐	☐

19. Indiquez approximativement le nombre de kilomètres que vous devez parcourir pour vous rendre chez le dentiste. __________ km

20. Visitez-vous ou avez-vous déjà visité régulièrement un orthodontiste avec votre enfant ?
☐ oui ☐ non

Si oui, combien de kilomètres deviez vous parcourir pour vous y rendre ? _______km

		Pas du tout	Un peu	Passable ment	Beaucoup
21.	Votre enfant a-t-il de la difficulté à mordre dans une pomme entière non pelée ?	☐	☐	☐	☐
22.	Votre enfant a-t-il de la difficulté à mastiquer des tranches de pomme ?	☐	☐	☐	☐
23.	Votre enfant a-t-il de la difficulté à mastiquer des morceaux de viande ?	☐	☐	☐	☐
24.	Pendant le repas, votre enfant a-t-il tendance à avoir des gaz ou des rots ?	☐	☐	☐	☐
25.	Normalement, votre enfant est-il constipé ?	☐	☐	☐	☐
26.	Pendant la journée, votre enfant a-t-il souvent la langue qui sort de sa bouche ?	☐	☐	☐	☐
27.	Est-ce que les gencives de votre enfant saignent facilement ?	☐	☐	☐	☐
28.	Votre enfant a-t-il mauvaise haleine ?	☐	☐	☐	☐
29.	Votre enfant grince-t-il des dents ?	☐	☐	☐	☐
30.	Est-ce que la nourriture sort de la bouche de votre enfant lorsqu'il mastique ?	☐	☐	☐	☐
31.	Votre enfant a-t-il facilement un haut le cœur ou a-t-il facilement envie de vomir lorsque quelque chose est placé dans sa bouche ?	☐	☐	☐	☐
32.	Votre enfant dort-il la bouche ouverte ?	☐	☐	☐	☐

		Oui	Non	Je ne sais pas
33.	Lorsque votre enfant était allaité ou nourri à la bouteille, avait-il de la difficulté à téter ?	☐	☐	☐
34.	Votre enfant a-t-il eu de la difficulté à passer de la nourriture en purée, à la nourriture solide ?	☐	☐	☐
35.	Est-ce que les dents de lait de votre enfant prennent du temps	☐	☐	☐

	à tomber ?			
36.	Votre enfant a-t-il déjà reçu un traitement dentaire ?	☐	☐	☐
		Oui	Non	Je ne sais pas
37.	Votre enfant a-t-il déjà pris des comprimés ou des gouttes de fluorure ?	☐	☐	☐
38.	Votre enfant a-t-il déjà reçu un traitement au fluorure chez le dentiste autre que des comprimés ou des gouttes ?	☐	☐	☐
39.	Votre enfant s'est-il déjà fait enlever une dent ?	☐	☐	☐
40.	Votre enfant a-t-il déjà eu un plombage ?	☐	☐	☐
41.	Votre enfant a-t-il déjà été endormi à l'aide d'une anesthésie générale avant de recevoir un traitement dentaire ?	☐	☐	☐
42.	Votre enfant a-t-il ou a-t-il déjà eu une prothèse dentaire, un pont ou une couronne ?	☐	☐	☐
43.	Votre enfant reçoit-il ou a-t-il déjà reçu un traitement d'orthodontie (traitement pour corriger la position des dents) ?	☐	☐	☐
44.	Les dents de votre enfant sont-elles brossées tous les jours ?	☐	☐	☐
45.	Votre enfant est-il capable de manger n'importe quelle sorte de nourriture ?	☐	☐	☐
46.	Votre enfant est-il capable de se laver les dents seul ?	☐	☐	☐
47.	Est-ce difficile d'évaluer si votre enfant souffre ?	☐	☐	☐
48.	Est-ce difficile de déterminer où votre enfant a mal ?	☐	☐	☐
49.	D'après ce que vous en savez, votre enfant a-t-il déjà souffert de problèmes dentaires ?	☐	☐	☐
50.	Votre enfant parle-t-il ?	☐	☐	☐
51.	Lorsque votre enfant parle, peut-il être compris par des gens en dehors de la famille immédiate ?	☐	☐	☐
52.	Votre enfant est-il capable de prononcer la lettre "T" (comme dans "table") ?	☐	☐	☐
53.	Votre enfant peut-il prononcer la lettre "g" (comme dans "grand-maman") ?	☐	☐	☐

		Très pauvre	Pauvre	Passable	Bonne	Excellente
54.	Comment qualifieriez-vous la santé générale de votre enfant ?	☐	☐	☐	☐	☐
55.	Comment qualifieriez-vous la santé buccale et dentaire de votre enfant ?	☐	☐	☐	☐	☐

10.13. English adult questionnaire: Control group

McGill University Down Syndrome Research Study

Subject ID ______________________

Date of questionnaire completion _______________ (dd/mm/yyyy)

1. At what level did you complete your formal education?
   - Elementary/High School ☐
   - College/CEGEP ☐
   - University ☐

2. How old are you? _______ years

3. What is your gender?
   - Male ☐
   - Female ☐

4. Which of the following best describes your ethnic background?
   - White ☐
   - Black ☐
   - Native North American ☐
   - Asian ☐
   - Other ☐ (please specify) ________________________

5. If you are suffering from any of the medical problems listed below, please note the frequency with which you see a doctor or a specialist concerning this problem.

	At least once a month	Several times a year	Once a year	Less than once a year	Not relevant
a) Heart problems	☐	☐	☐	☐	☐
b) Immunological problems	☐	☐	☐	☐	☐
c) Ear infections, tonsilitis, etc..	☐	☐	☐	☐	☐
d) Speech problems	☐	☐	☐	☐	☐
e) Visual problems	☐	☐	☐	☐	☐
f) Orofacial or dental problems	☐	☐	☐	☐	☐
g) Other ☐ (please specify) ______________________	☐	☐	☐	☐	☐

	Not at all	A little	Quite a lot	Very much
6. Do you grind your teeth?	☐	☐	☐	☐

		Yes	No	I don't know
7.	Have you ever taken fluoride tablets or drops?	☐	☐	☐
8.	Have you ever received any form of fluoride treatment at the dentist?	☐	☐	☐
9.	Have you ever had a tooth extracted?	☐	☐	☐
10.	Have you ever had a filling?	☐	☐	☐
11.	Have you ever been put to sleep (with a general anaesthetic) for a dental procedure?	☐	☐	☐
12.	Does your child have, or has he/she ever had, a denture, a bridge, or a crown?	☐	☐	☐
13.	Have you in the past, or currently undergoing orthodontic treatment (treatment to correct tooth position)?	☐	☐	☐
14.	Do you brush your teeth everyday?	☐	☐	☐

	Very Poor	Poor	Reasonable	Good	Excellent
15. How would you rate your overall oral/dental health?	☐	☐	☐	☐	☐

10.14. French adult questionnaire: Control group

Université McGill: Un projet de recherche chez les individus trisomiques

ID du sujet ______________________

Date _______________ (dd/mm/aaaa)

---

1. Quel est votre niveau de scolarité?
   - Primaire/Secondaire ☐
   - Collège/CEGEP ☐
   - Université ☐

2. Quel âge avez vous? _________ans

3. Quel est votre sexe?
   Masculin ☐ Féminin ☐

4. Lequel des choix suivants décrit le mieux votre origine ethnique?
   - Blanc ☐
   - Noir ☐
   - Amérindien ☐
   - Asiatique ☐
   - Autre ☐ (spécifiez s.v.p.) _________________________

5. Si vous souffrez d'un ou de plusieurs problèmes mentionnés dans la liste suivante, veuillez s.v.p. indiquer le nombre de fois que ces problèmes vous obligent à consulter un médecin ou un spécialiste.

	Au moins une fois par mois	Plusieurs fois par année	Une fois par année	Moins d'une fois par année	Ne s'applique pas
a) Problèmes cardiaques	☐	☐	☐	☐	☐
b) Problèmes immunitaires (ex: leucémie)	☐	☐	☐	☐	☐
c) Infections aux oreilles, amygdalite etc.	☐	☐	☐	☐	☐
d) Problèmes de langage	☐	☐	☐	☐	☐
e) Problèmes visuals	☐	☐	☐	☐	☐
f) Problèmes bucco-dentaires	☐	☐	☐	☐	☐
g) Autre ☐ (spécifiez s.v.p.) ______________________	☐	☐	☐	☐	☐

		Pas du tout	Un peu	Passablement	Beaucoup
6.	Est-ce que vous grincez des dents ?	☐	☐	☐	☐

		Oui	Non	Je ne sais pas
7.	Est-ce que vous avez déjà pris des comprimés ou des gouttes de fluorure ?	☐	☐	☐
8.	Est-ce que vous avez déjà reçu un traitement au fluorure chez le dentiste autre que des comprimés ou des gouttes?	☐	☐	☐
9.	Est-ce que vous avez déjà fait enlever une dent?	☐	☐	☐
10.	Est-ce que vous avez déjà eu un plombage?	☐	☐	☐
11.	Est-ce que vous avez déjà été endormi à l'aide d'une anesthésie générale avant de recevoir un traitement dentaire?	☐	☐	☐
12.	Est-ce que vous avez déjà eu une prothèse dentaire, un pont ou une couronne?	☐	☐	☐
13.	Est-ce que vous recevez ou avez déjà reçu un traitement d'orthodontie (traitement pour corriger la position des dents)?	☐	☐	☐
14.	Est-ce que vos dents sont brossées tous les jours?	☐	☐	☐

	Très pauvre	Pauvre	Passable	Bonne	Excellente
15. Comment qualifieriez-vous votre santé buccale et dentaire?	☐	☐	☐	☐	☐

10.15. English parental questionnaire: Control group

McGill University Down Syndrome Research Study

Subject ID ______________________

Date of questionnaire completion _______________ (dd/mm/yyyy)

1. Are you your child's...
   - Mother ☐
   - Father ☐
   - Brother/sister ☐
   - Guardian ☐

2. At what level did you (the parent/guardian) finish your formal education?
   - Elementary/High School ☐
   - College/CEGEP ☐
   - University ☐

3. How old is your child? _______ years

4. What is your child's gender?
   - Male ☐
   - Female ☐

5. Which of the following best describes your child's ethnic background?
   - White ☐
   - Black ☐
   - Native North American ☐
   - Asian ☐
   - Other ☐ (please specify) ________________________

6. If your child suffers from any of the medical problems listed below, please note the frequency with which he/she sees a doctor or a specialist concerning this problem.

	At least once a month	Several times a year	Once a year	Less than once a year	Not relevant
a) Heart problems	☐	☐	☐	☐	☐
b) Immunological problems	☐	☐	☐	☐	☐
c) Ear infections, tonsilitis, etc..	☐	☐	☐	☐	☐
d) Speech problems	☐	☐	☐	☐	☐
e) Visual problems	☐	☐	☐	☐	☐
f) Orofacial or dental problems	☐	☐	☐	☐	☐
g) Other ☐ (please specify) ______________________	☐	☐	☐	☐	☐

		Not at all	A little	Quite a lot	Very much
7.	Does your child grind his/her teeth?	☐	☐	☐	☐

		Yes	No	I don't know
8.	Has your child ever taken fluoride tablets or drops?	☐	☐	☐
9.	Has your child ever received any form of fluoride treatment at the dentist?	☐	☐	☐
10.	Has your child ever had a tooth extracted?	☐	☐	☐
11.	Has your child ever had a filling?	☐	☐	☐
12.	Has your child ever been put to sleep (with a general anaesthetic) for a dental procedure?	☐	☐	☐
13.	Does your child have, or has he/she ever had, a denture, a bridge, or a crown?	☐	☐	☐
14.	Is your child undergoing, or has he/she ever undergone, orthodontic treatment (treatment to correct tooth position)?	☐	☐	☐
15.	Are your child's teeth brushed daily (by him/her or by somebody else)?	☐	☐	☐

	Very Poor	Poor	Reasonable	Good	Excellent
16. How would you describe the oral/dental health of your child?	☐	☐	☐	☐	☐

10.16. French parental questionnaire: Control group

Université McGill: Un projet de recherche chez les individus trisomiques

ID du sujet ______________________

Date _______________ (dd/mm/aaaa)

1. Quel est votre lien de parenté avec votre enfant?
   - Mère ☐
   - Père ☐
   - Frère/Soeur ☐
   - Tuteur ☐

2. Quel est votre niveau de scolarité?
   - Primaire/Secondaire ☐
   - Collège/CEGEP ☐
   - Université ☐

3. Quel âge a votre enfant? _________ans

4. Quel est le sexe de votre enfant?
   Masculin ☐ Féminin ☐

5. Lequel des choix suivants décrit le mieux l'origine ethnique de votre enfant?
   - Blanc ☐
   - Noir ☐
   - Amérindien ☐
   - Asiatique ☐
   - Autre ☐ (spécifiez s.v.p.) _________________________

6. Si votre enfant souffre d'un ou de plusieurs problèmes mentionnés dans la liste suivante, veuillez s.v.p. indiquer le nombre de fois que ces problèmes l'obligent à consulter un médecin ou un spécialiste.

	Au moins une fois par mois	Plusieurs fois par année	Une fois par année	Moins d'une fois par année	Ne s'applique pas
a) Problèmes cardiaques	☐	☐	☐	☐	☐
b) Problèmes immunitaires (ex: leucémie)	☐	☐	☐	☐	☐
c) Infections aux oreilles, amygdalite etc.	☐	☐	☐	☐	☐
d) Problèmes de langage	☐	☐	☐	☐	☐
e) Problèmes visuels	☐	☐	☐	☐	☐
f) Problèmes bucco-dentaires	☐	☐	☐	☐	☐
g) Autre ☐ (spécifiez s.v.p.) _____________________	☐	☐	☐	☐	☐

		Pas du tout	Un peu	Passablement	Beaucoup
7.	Votre enfant grince-t-il des dents?	☐	☐	☐	☐

		Oui	Non	Je ne sais pas
8.	Votre enfant a-t-il déjà pris des comprimés ou des gouttes de fluorure?	☐	☐	☐
9.	Votre enfant a-t-il déjà reçu un traitement au fluorure chez le dentiste autre que des comprimés ou des gouttes?	☐	☐	☐
10.	Votre enfant s'est-il déjà fait enlever une dent?	☐	☐	☐
11.	Votre enfant a-t-il déjà eu un plombage?	☐	☐	☐
12.	Votre enfant a-t-il déjà été endormi à l'aide d'une anesthésie générale avant de recevoir un traitement dentaire?	☐	☐	☐
13.	Votre enfant a-t-il actuellement ou a-t-il déjà eu une prothèse dentaire, un pont ou une couronne?	☐	☐	☐
14.	Votre enfant reçoit-il ou a-t-il déjà reçu un traitement d'orthodontie (traitement pour corriger la position des dents)?	☐	☐	☐
15.	Les dents de votre enfant sont-elles brossées tous les jours?	☐	☐	☐

	Très pauvre	Pauvre	Passable	Bonne	Excellente
16. Comment qualifieriez-vous la santé buccale et dentaire de votre enfant?	☐	☐	☐	☐	☐