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A paired comparison of dental care in Canadians with Down syndrome and their siblings without Down syndrome

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Abstract – *Objective:* To test the hypothesis that, controlling for age, Canadians with Down syndrome (DS) have dental care that is different to that of their siblings without DS. *Methods:* A cross-sectional survey of parents of children with DS among members of the Canadian Down Syndrome Society (CDSS), using a validated questionnaire. Parents were asked to complete two versions of the questionnaire: one for their child with DS and another for the sibling closest in age without DS. A total of 2327 questionnaires were distributed; 1221 questionnaires for people with DS and 950 for siblings without DS were returned. A paired analysis (McNemar test) of dental care indicators was performed on data from 938 family pairs, stratifying for age. *Results:* For all the dental care indicators and age groups, many respondents indicated the same behaviours or experiences in their child with DS and a sibling without DS. However, depending on the particular form of dental care and the age group, 0–47% of families reported discordant dental care experiences for their child with DS and a sibling without DS. The greatest differences were observed for yearly consults ($P = 0.029$), restorations ($P < 0.001$), fluoride therapy ($P = 0.013$) and extractions ($P = 0.029$). *Conclusion:* These observations suggest that compared to their siblings without DS, Canadians with DS are receiving different dental care.

Key words: access; Canada; dental care; dental treatment; Down syndrome

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Down syndrome (DS) is an autosomal chromosomal anomaly resulting from trisomy of all, or a critical part, of chromosome 21 (1). There are no birth registries of people with DS in Canada, but estimates of the incidence in USA and Europe vary from approximately 9.0–12.5 per 10 000 live births (1–6). However, quite large variations exist for different American states and within different ethnic groups (4).

Many of the pathological and physiological characteristics, including the orofacial problems of DS, have been described, and it is evident that this group suffers from very high levels of orofacial disease and other problems (7). If this is the case, then one important avenue of research concerning people with DS is their access to dental care. The dental care of people with DS is different to that of the non-DS population at several levels. The provision and/or supervision of oral hygiene by parents has been reported to be worse than that for non-DS controls (8, 9), and this is exacerbated if the individuals with DS are institutionalised where very poor oral hygiene care has been reported (10–12). Dental consultation patterns vary by country. Studies from France and Denmark reported that children with DS were more likely to visit a dentist yearly than their non-DS siblings (9) or other non-DS groups (13), while a study in the UK found that the inverse was the case, although this study included subjects with handicaps other than those with DS (14). There are also reports of people with DS having problems finding a dentist to treat them (9, 15, 16), and many

have to use hospital care (15, 17). Furthermore, once they have accessed some form of dental care, people with DS are less likely to receive preventive and restorative care and more likely to be treated with dental extraction, than people without DS (9, 12, 15). A number of publications have discussed the ambivalent attitudes of dental professionals towards people with DS and other mental and physical handicaps (15, 17, 18), and there has been suggestion of the need for dentists specially trained to care for such groups (16, 19).

In Canada, we have no information concerning either the prevalence of oral health problems experienced by, or the type of dental care provided for people with developmental disabilities. In view of this observation, we performed a study to investigate the provision of dental care to people with DS in Canada. More specifically, the objective of this study was to test the hypothesis that, controlling for age, Canadians with DS receive dental care that is different to that received by their siblings without DS. A number of indicators of dental care were used to test this overall hypothesis.

Methodology

Study design and sample

A population-based, cross-sectional survey design was used, with the Canadian Down Syndrome Society (CDSS) membership as the sample source. The CDSS membership at the time of the study was 2327 families with one or more children with DS. Data were gathered through the use of a questionnaire. A cover letter, two copies of the questionnaire and a postage-paid reply envelope were distributed by the CDSS to all their members. Parents were asked to complete the two-labelled and colour-coded copies of the questionnaire: one with respect to their child with DS and the second concerning the sibling without DS closest in age. The cover letter requested that the same parent should complete both questionnaires, although this was not verifiable. A second mailing was performed to those who had not responded initially, 1 month following the first mailing. In total, 1221 questionnaires concerning people with DS and 950 concerning their siblings were returned, representing a 52.5% response rate for the children with DS. In view of the analytic need to make paired comparisons, only data concerning the 950 people with DS with a sibling questionnaire were potentially usable. Of these 950 pairs of questionnaires, 938 had all the necessary variables completed in the questionnaires. Analyses were performed on these 938 pairs of questionnaires. Prior to it's starting, the study was reviewed and approved by the McGill University IRB and by the executive committee of the CDSS. Consent was implicit in parents agreeing or not to complete the questionnaires. All questionnaire distribution and collection were performed through the CDSS to guard subject anonymity.

Variables

Among other background questions concerning age, gender, residential location, daily activities and general health status of the subject of the questionnaire (i.e. the child and not the parent), the questionnaire contained eight questions concerning home and professional dental care. These questions were as follows: (i) Does your child visit the dentist yearly? (ii) Has your child ever taken fluoride tablets or drops? (iii) Has your child ever received any form of fluoride treatment at the dentist? (iv) Has your child ever had a tooth extracted? (v) Has your child ever had a filling? (vi) Has your child ever been put to sleep (had a general anaesthetic) for a dental procedure? (vii) Does your child have, or has he/she ever had a crown, a bridge or a denture? and viii) Is your child undergoing, or has your child ever undergone orthodontic treatment (treatment to correct tooth position)? The response to all questions was 'yes', 'no' or 'I don't know'.

Statistical analyses

Following descriptive statistics, age-stratified paired bivariate analyses were performed to investigate the relationship between DS status and the dependent, dental care variables in different age strata. These paired bivariate analyses were performed using the McNemar test. For any question, there are four possible combinations of responses for each set of paired data: (i) the response for both the individual with DS and the sibling was 'no'; (ii) the response for both the individual with DS and the sibling was 'yes'; (iii) the response for the individual with DS was 'yes' and for the sibling was 'no'; and (iv) the response for the individual with DS was 'no' and for the sibling was 'yes'. As we are interested in finding situations where the behaviour of the family pairs is different, the analysis focuses on the discordant (yes/no versus no/yes) pairs only. The McNemar test compares the number of discordant pairs in the 2×2 contingency tables testing the hypothesis that the proportion of 'yes/no' is the same as that of 'no/yes'. For example, in our data set, it tests the

hypothesis that the number of family pairs in which the parent reported that the child with DS does consult a dentist yearly while the sibling does not, is the same as vice versa. As usual, a *P*-value of less than 0.05 suggests that the proportions are statistically different. Pairs were excluded from the analyses if response for one or both of the subjects was 'I don't know'.

The matched nature of this data set considerably reduces the number of possible correlates of differences in proportions of people receiving or not receiving the various forms of dental care beyond DS status. However, it is conceivable that age, gender, place of residence, daily activity (as an indicator of level of disability), dental and other health status indicators are different within the pairs and that they are related to the dental care variables. The effect of age was demonstrated through the age-stratified McNemar analyses described above. Clinical dental status data were not collected. However, data concerning gender, place of residence, daily activity and health status were collected, enabling them to be controlled for in analysing the relationship between differences in dental care by DS status. Principal daily activity was classified as 'at home', 'at normal school', 'at specialised school/day centre', 'working in the community' or 'working in a protected environment'. Those staying at home, attending a specialised school or working in a protected environment were considered to have a more severe disability than those attending a normal school or working in the community. Health status was assessed through questions concerning the frequency of visits to health care professionals for selected health problems (heart, immunological, ENT and speech). Those children whose parents reported such consultations more frequently than once a year were categorised as having a problem. Conditional logistic regression analysis was performed to assess the association between DS status and each of the dental care variables, controlling for age, gender, place of residence and health status.

Results

Descriptive statistics for the people with DS and their siblings are shown in Table 1. There was an even mix of genders in both groups, and the large majority of sample subjects were children, with the mean age for the DS group being 10.7 years (SD 8.0) and that for the sibling group being 12.0 years (SD 8.7). In the paired analysis, the mean age difference between pairs was 3.4 years. As their normal daily activity, a large majority of people in both the groups were living at home and going to a normal school. Significant minorities of the group with DS had heart and immunological problems, while large proportions had ENT and speech problems. Among their siblings, 11.1% had ENT problems, but otherwise the prevalence of health problems was very low.

The results of the age-stratified paired analyses for each of the eight dental care variables are demonstrated in Figs 1–8. Different patterns emerge with each of the variables. For the yearly consultation (Fig. 1), while in the youngest age group, there are more children with DS not consulting while their sibling does than vice versa (32% vs. 7%; $P < 0.001$), for all other age groups, there is a strong trend or a significant difference in the opposite direction with children with DS more likely to consult while their sibling does not than vice versa ($P = 0.08$–0.001). It is important to note, however, that for all age groups, the majority of families report the same behaviour (2–12% both not consulting and 41–87% both consulting) for both children in the DS/sibling pairs. With home- and professionally applied fluorides (Figs 2 and 3), the same pattern emerges across age groups. For the four younger age groups (0–18 years), significantly more families report their child with DS not receiving either form of fluoride, while the sibling does than vice versa. With both fluoride formats, the adult group shows no difference.

The relationship of DS status and having experienced a tooth extraction (Fig. 4) differs with age. In the youngest two groups (0–6 years), children with DS are more likely not to have had a tooth extraction while their sibling has, rather than vice versa. There is no *P*-value for the 0–3-year-old age group because two of the boxes have zero counts, so a McNemar test cannot be performed. Nevertheless, the comparison of 9% of pairs in which the child with DS has not had an extraction while their sibling has versus 0% of pairs in which the child with DS has had an extraction while the sibling has not is strongly suggestive of an important difference. In the 7–18-year-old age groups, however, the intrafamily extraction–experience differences are in the opposite direction, with the children with DS having an extraction while their sibling does not, being significantly more common than the reciprocal.

The relationship between DS status and receipt of a restoration (Fig. 5) is the only dental care variable wherein the association does not change with age. In all age groups, children with DS are more likely not

Table 1. Sample descriptive statistics

Variable	Category	DS group *N* (%)	Siblings *N* (%)
Gender	Male	491 (52.3)	464 (49.5)
	Female	447 (47.7)	474 (50.5)
Age (years)	0–3	143 (15.2)	96 (10.2)
	4–6	161 (17.2)	169 (18.0)
	7–12	350 (37.3)	314 (33.5)
	13–18	178 (19.0)	215 (22.9)
	19+	104 (11.1)	142 (15.1)
Residence	With parents	895 (95.4)	822 (87.6)
	Elsewhere	39 (4.6)	110 (12.4)
Daily activities	At home	91 (9.7)	97 (10.3)
	At normal school	593 (63.2)	695 (74.1)
	At specialised school or day centre	198 (21.1)	38 (4.1)
	Working in the community	27 (2.9)	96 (10.2)
	Working in a protected environment	20 (2.1)	1 (0.1)
Health status	Heart problems	147 (15.6)	2 (0.2)
	Immunological problems	114 (12.1)	15 (1.5)
	ENT problems	434 (46.3)	104 (11.1)
	Speech problems	544 (58.0)	24 (2.6)
Yearly consult	Yes	780 (86.7)	724 (80.4)
	No	120 (13.3)	176 (19.6)
Fluoride tabs/drops	Yes	265 (30.4)	333 (38.2)
	No	606 (69.6)	538 (61.8)
Professional fluoride	Yes	502 (57.2)	662 (75.5)
	No	375 (42.8)	215 (24.5)
Extraction	Yes	333 (36.4)	283 (30.9)
	No	582 (63.6)	632 (69.1)
Restoration	Yes	237 (26.0)	460 (50.4)
	No	675 (74.0)	452 (49.6)
GA	Yes	215 (23.4)	83 (9.1)
	No	702 (76.6)	834 (90.9)
Crown/bridge/denture	Yes	46 (5.0)	37 (4.0)
	No	879 (95.0)	888 (96.0)
Orthodontic treatment	Yes	132 (14.3)	207 (22.5)
	No	788 (85.7)	713 (77.5)

to have had a restoration while their sibling has ($P < 0.001$ for all groups). With respect to general anaesthetic experience (Fig. 6), while the two youngest groups show no difference, children with DS in the older age groups (>6 years) are more likely to have had this experience while their sibling has not than vice versa ($P = 0.038$–001). There was no association between the receipt of complex restorative/

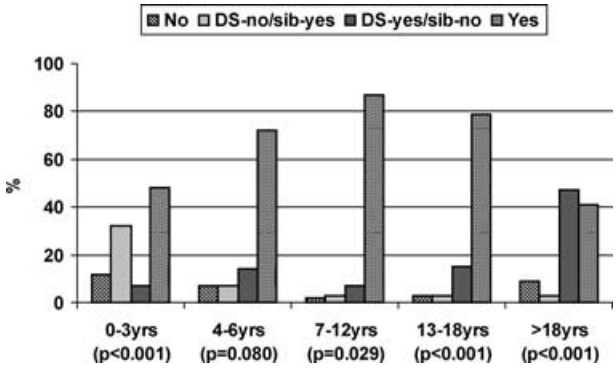

Fig. 1. Yearly consultation with a dentist.

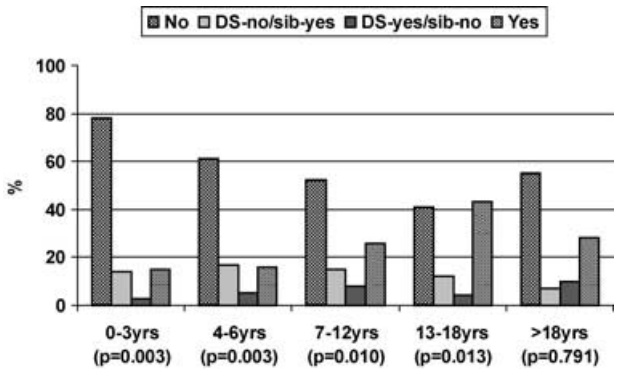

Fig. 2. Ever use of fluoride tablets/drops.

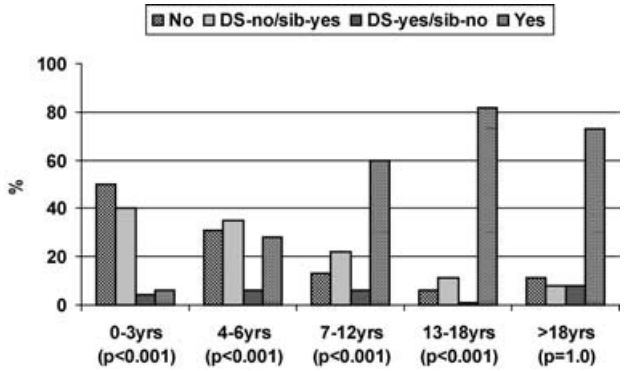

Fig. 3. Ever professional fluoride use.

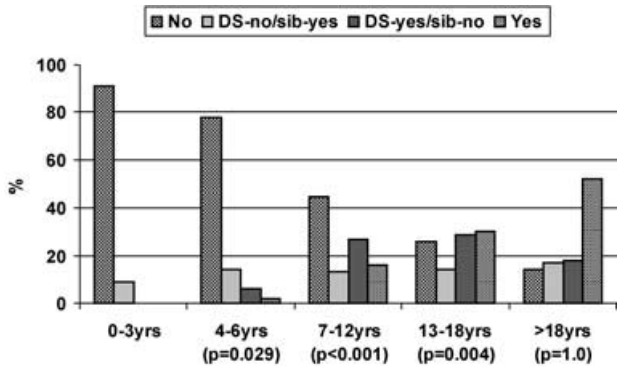

Fig. 4. Ever had a tooth extracted.

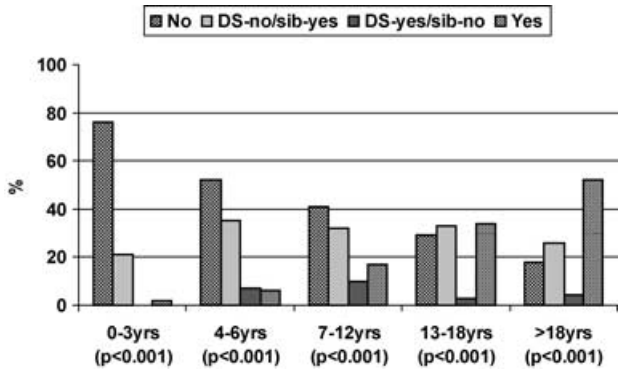

Fig. 5. Ever had a filling.

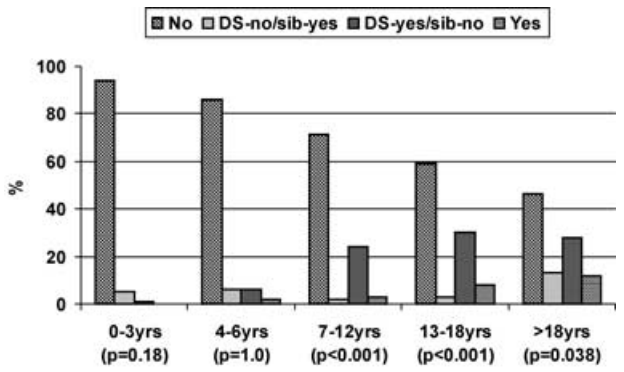

Fig. 6. Ever had a general anaesthetic.

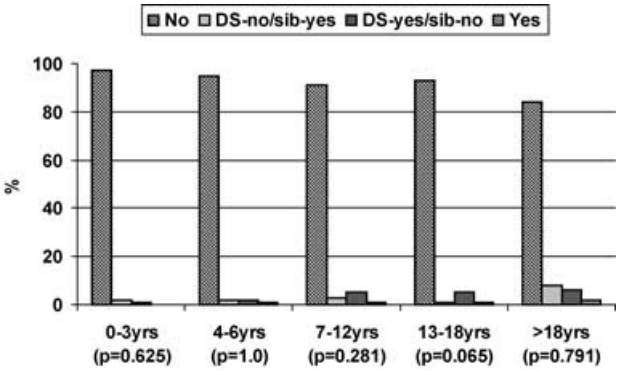

Fig. 7. Ever had a crown, bridge or denture.

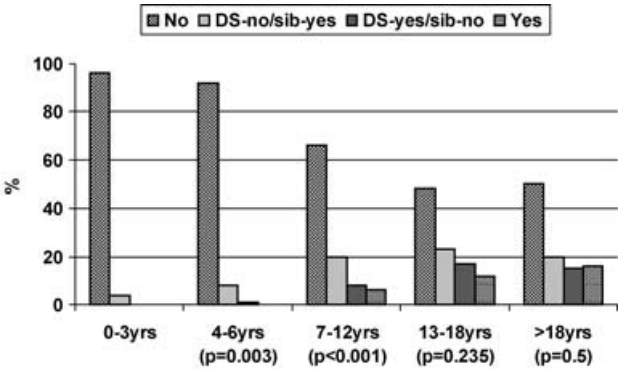

Fig. 8. Ever had orthodontic treatment.

prosthetic treatment and DS status (Fig. 7) for all age groups. Finally, for children aged 4–12 years, families in which the child with DS had not received orthodontic treatment while their sibling had received such treatment were more frequent than the reciprocal arrangement ($P = 0.003$); however, no differences were evident in the other age groups (Fig. 8).

As stated in the methodology, a multivariate analysis of the association between DS status and dental care variables, controlling for gender, place of residence daily activity and health status was performed. However, none of these variables was associated with any of the dental care variables at a bivariate level of analysis and none of the aforementioned age-stratified associations between DS status and dental care was altered through the use of multivariate modelling. We are therefore not showing the results of these analyses.

Discussion

This study was designed to test the hypothesis that selected aspects of dental care differ between people

with DS and a sibling, when controlling for age. The findings of our study support this hypothesis for the majority of the eight dental care indicators used, although the direction of the differences differs with the dental care variable and sometimes with age as well.

Having observed these differences, it is important to note that the analyses we have performed focus on discordant pairs, and for the large majority of age-stratified comparisons, there are more families with concordant than discordant behaviours or experiences. For example, depending on the age, 41–87% of both siblings consult a dentist yearly, 41–78% of both siblings have never had fluoride tablets or drops and 48–96% of both siblings have not had an orthodontic treatment. It is also important to note that this study has several shortcomings that limit our ability to make inferences from the analytic findings. Most notable are the sample source and the response rate. The data came from members of the CDSS, who probably are the better educated and most motivated of Canadian parents of children with DS. Although there is no formal record, the CDSS estimates that there are 42 000 people with DS in Canada (personal communication). The questionnaires were sent to their members (2327) and the response rate was 52.5%, although for the purposes of our analysis, we only used 938 paired data sets, i.e. 40.3% of the questionnaires were distributed. Beyond these important sample biases, it is important to note that the dental care variables were crude indicators of access and that the majority of them referred to ever having received a particular form of dental care rather than having received it during the past year. This means that differences observed by age do not necessarily mean that the different treatments occurred while the children were in that age category. Rather, it means that the differences were accumulated by the age category. It also explains why the differences with fluoride therapies, tooth extraction and orthodontic therapy disappear with increasing age, while the one question concerning a yearly rather than an ever experience (yearly consultation) remains different into adulthood. However, this point also emphasises the apparently lifelong difference in restoration experience observed in our sample.

Another important point is that the age groups were based upon the age of the child with DS, and that the mean age difference between the child and their sibling was 3.4 years. Some form of age difference was, however, inevitable unless we used a design with twins only, in which there was a child with DS and a twin without. A few such twins (four twins) did exist in our sample, but they were not enough to study our hypothesis. The final limitation concerns the nature of the daily activity and health status indicators and the lack of dental status indicator. The indicators we used for the former two variables were crude, which may explain why they had no independent effect. However, as they are most relevant to the group with DS, their use would be more important in an analysis of different dental care within that group alone rather than a comparison with non-DS siblings. The lack of a dental status indicator is, however, an important limitation in our understanding of the findings. If we had collected dental status data, we would be better able to address the question of whether observed dental care differences between the two groups were because of their DS or because of their dental status.

Having outlined these limitations, it is also important to note that although the sample has its aforementioned biases, it is a large sample with a study design and analytic technique (paired analyses) that controls very well for the majority of variables that are recognised determinants of access to dental care in children, e.g. family socioeconomic status, dental insurance coverage, geographic location and parental education levels. All these variables were matched.

Looking at the study findings concerning caries management specifically, our results fit the popularly held belief that people with DS have lower rates of caries than people without DS (20–22). In our sample, where there was family discordance, children with DS not receiving fluoride or restorative therapy, while a sibling did, was more common than the opposite case. However, bearing in mind the limitations of the research claiming lower caries rates in people with DS (7, 23), it remains to be demonstrated with appropriately designed research whether this difference in caries management experience is because of differences in disease rates and/or management strategies in people with DS and those without. It is interesting to note, however, that the findings of our study concerning yearly dental consultations are in agreement with similar studies in Europe (9, 13). Further research is required to explain this increasingly evident dental care dynamic wherein people with DS consult the dentist more often than those without DS, but receive less caries-preventive and restorative care and more extractions.

The differences in dental extraction experiences observed in our study are difficult to explain. There are numerous possible reasons for dental extractions. It is, however, interesting to note than where there are discordant experiences within families, siblings having an extraction, while the child with DS did not, was more common than vice versa with the 0–6-year-old age groups, but the inverse (pairs in which the child with DS had experienced an extraction while the sibling had not was more common than the reciprocal) was true for 7–18-year-old age groups. This suggests that for some reason, there is a dramatic change in relative extraction experience during the transition from the 0–6 to 7–18-year-old age groups. This could be related to the mixed dentition stage in the children with DS and the well-recognised delayed deciduous tooth exfoliation and/or permanent tooth eruption experienced by this group (7). Finally, it is not surprising to see that where there is discordance within families, the child with DS having experienced a general anaesthetic for a dental procedure, while their sibling has not, is more common than the reciprocal situation. This finding is in agreement with the work from Europe showing the relatively common need to use such anaesthesia to provide to people with DS dental care (15, 17).

In conclusion, this paired analysis of within-family differences in dental care behaviours and experiences for people with DS and non-DS siblings demonstrates that where there are differences, people with DS are more likely to consult a dentist yearly but less likely to receive caries-preventive and restorative care, and more likely to have had a dental extraction.

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