MªA Mayoral Trias*, J. Llopis-Perez**, A. Puigdollers Pérez***

Department of Orthodontics, School of Dentistry, Universitat Internacional de Catalunya, Sant Cugat del Vallés, Barcelona, Spain
**Associate professor*
***Statistics Consultant*
****Associate Professor and Department Chair*

e-mail: dramayoral@clinicamayoral.com

Comparative study of dental anomalies assessed with panoramic radiographs of Down syndrome and non-Down syndrome patients

ABSTRACT

Aim *The aim of this study was to compare the prevalence of dental anomalies from panoramic radiographs of age-matched individuals with and without Down Syndrome (DS).*
Materials and methods *Study Design: This is a retrospective cross-sectional study. A group of 41 patients (19 female and 22 male) with Down Syndrome (DS), mean age 10.6 ±1.4 and a control group of 42 non- DS patients (26 female and 16 male), mean age 11.1 ±1.3 were studied. Methods: This study examined the medical history and a panoramic radiograph of each patient. The dental anomalies studied were agenesis of permanent teeth (except third molars), size and shape maxillary lateral anomalies and maxillary canine eruption path anomalies. Statistics: The groups were compared using Mann-Whitney and Wilcoxon non-parametric tests (p<0.05). Rho Spearman correlation coefficient was applied for associations.*
Results *Agenesis of one permanent tooth was found in 73.17% of DS subjects and two or more permanent teeth in more than 50% (p<0.001). Maxillary lateral incisor was the most frequently absent tooth followed by mandibular second premolar, mandibular lateral incisor, maxillary second premolar and mandibular central incisor. No significant differences were detected between maxilla and mandible on either side. No differences in gender were observed. Significant differences were found for size and shape anomalies of maxillary lateral incisors, as well as for canine eruption anomalies (p<0.05). No gender differences were observed for either variable. No association was found between these two variables in the DS group.*
Conclusions *More dental anomalies were present in the DS group than in the control group, which implied that DS patients need periodical dental and orthodontic supervision so as to prevent or control subsequent oral problems.*

Keywords *Agenesis of teeth; Canine eruption path anomalies; Down Syndrome; Size and shape incisor anomalies.*

Introduction

Down Syndrome (DS), also known as trisomy 21 or G, is the most prevalent genetic malformation and affects one out of 700 births. This malformation was first described in 1866 by the English physician Langdon Down. In 1959 Lejeune and Turpin identified numerical chromosomal alterations characterised by trisomy 21 in DS subjects. The principal features of DS consist of a peculiar phenotype, generalised muscular hypotony, intellectual discapacity and growth retardation. Several systemic cardiovascular, neurological, gastrointestinal, ophthalmological, otolaryngological, musculoskeletal, hematopoietic, endocrynologic and dermatologic anomalies may be present [Desai, 1997]. Dental anomalies include hypodontia with missing lateral, premolar and wisdom teeth; peg-shaped laterals; microdontia; canine eruption path anomalies and delayed and irregular dental eruption in temporary and permanent dentitions. Midface hypoplasia and maxillary transverse micrognatism are frequently present predisposing factors to a Class III malocclusion with a concave profile and uni- or bilateral crossbite. Anterior open bite may also be found concurrent with lingual protrusion [Musich, 2006; Menendez Nuñez et al., 1992].

Many conditions connected to DS have been improved thanks to special health programmes, early stimulation, developed since the 80s, and growing awareness by the general public of intellectual disability. Previous life expectancy of 30 to 40 years has been radically extended to between 60 and 70 years. Given that DS patients require ever more dental and orthodontic care, in addition to the little research in this area, we believe it is necessary to gain more in-depth knowledge of dentomaxillofacial anomalies in DS subjects in order to optimise and contribute to their general health improvement.

This study focuses on the prevalence of dental anomalies (agenesis, size and shape of upper lateral incisors and upper canine eruption anomalies) assessed with panoramic radiographs in a group of DS patients compared to an age-matched group of non DS patients.

	DS group	Control group
N	41	42
female	19	26
male	22	16
Mean age	10.6 ±1.4	11.1 ±1.3

TABLE 1 Sample characteristics.

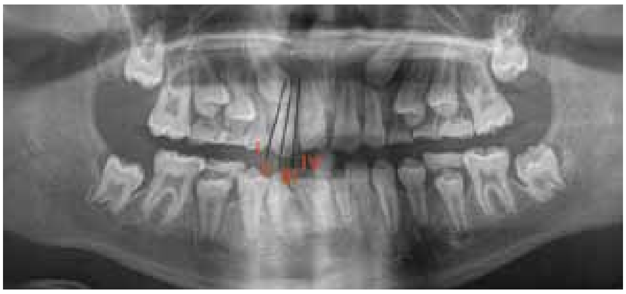

FIG 1 Lindauer et al. definition of canine eruption sectors used in this study.

Materials and Methods

Material

A group of 41 DS subjects was selected consecutively from the Dentistry Department of the Medical Centre of the Catalan Down Syndrome Foundation. An age-matched control group was consecutively selected from the Orthodontic Department of the Dental School at the Universitat Internacional de Catalunya. The inclusion criteria were: patients aged between 9 and 13 years of age, panoramic radiographs of sufficient quality, and no extraction or orthodontic treatment prior to the study. Table 1 describes the two groups.

Methods

The following variables were analysed from medical histories and panoramic radiographs: agenesis of permanent teeth, except third molars, anomalies in size and of shape maxillary lateral incisor such as microdontia, peg-shaped form, and maxillary canine eruption path anomalies. Alterations in canine eruption path were assessed using a modified method of Ericsson and Kurol [1986] and proposed by Lindauer et al. [1992]. According to this method, four sectors are defined on the basis of the position of the unerupted canine cusp tip with respect to the lateral incisor root (Fig. 1). Sector I is the area distal to a line tangent to the distal height of the contour of the lateral incisor crown and root; sector II is mesial to sector I, but distal to a line bisecting the mesiodistal dimension of the lateral incisor along the axis of the tooth; sector III is mesial to sector II, but distal to a line tangent to the mesial height of the contour of the lateral incisor crown and root; sector IV includes all areas mesial to sector III. The maxillary central incisor is taken as a reference when the maxillary lateral incisor is missing. Alteration of canine eruption path occurs when the canine is located in sectors II, III or IV and/or in case of uneruption with complete root development (impaction).

All measurements were performed by one observer. To determine the reliability of the measurements 10 randomly selected subjects from each group were examined twice. An intraclass correlation coefficient analysis was applied.

Statistical analysis

Data were entered in MS Excel and analysed using SPSS statistical software. The Shapiro-Wilk test for $n<50$ was applied to analyze quantitative variables. Significance level was set at $p< 0.05$ for all tests. Non-parametric tests were applied to data that did not follow a normal distribution. The power of the sample was calculated (99%). The mean values and standard deviations were computed for all variables. The two groups were compared with Mann-Whitney and Wilcoxon W non-parametric tests. Spearman Rho was used to assess correlation between size and shape lateral anomalies and upper canine eruption path anomalies.

Results

Agenesis (hypodontia)

The frequency of hypodontia for permanent teeth

	DS		Non DS		Significance
	Absolute Frequency	Relative Frequency	Absolute Frequency	Relative Frequency	P
Agenesis	30	73.17%	3	7.14%	P<0.0001*
Size/Shape Maxillary Lateral Incisor Anomalies	10	24.4%	1	2.4%	P=0.0037*
Upper Canine Eruption Path Anomalies	29	35.4%	11	13.1%	P=0.0008*

TABLE 2 The frequency and comparison between groups.

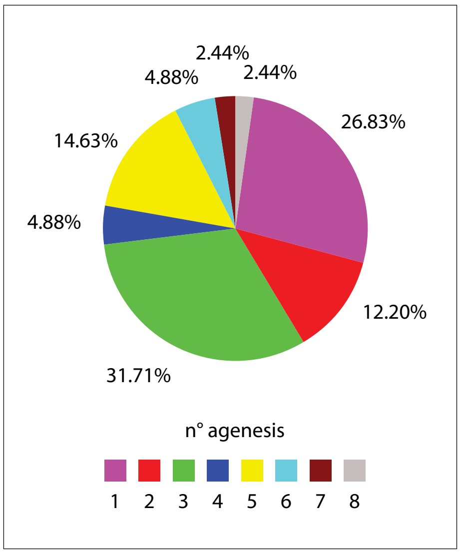

FIG 2 Agenesis percentage in DS group N= 41.

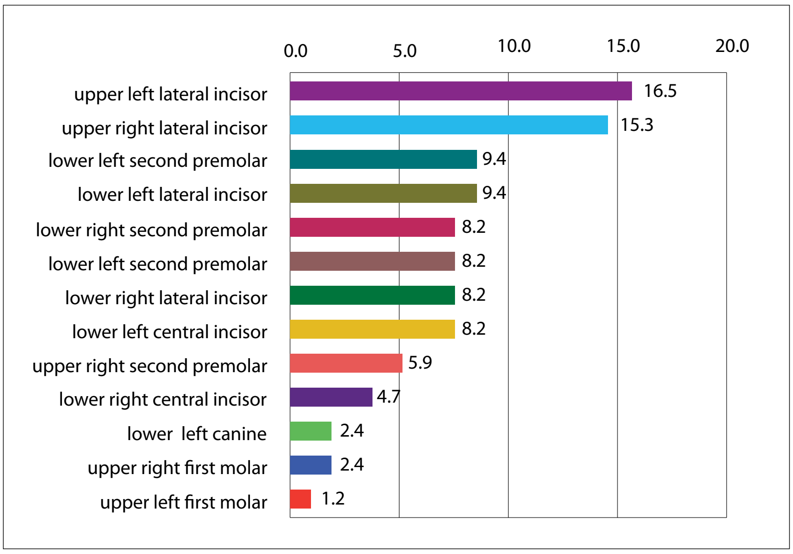

FIG 3 Distribution of agenesis by tooth type in DS group

was 73.17% in subjects with DS, whereas it was 7.14% for the control group. The difference between groups was statistically significant (p<0.000). Table 2 shows comparisons between the two groups. Two or more teeth were missing in more than 50% of the DS group. The average number of agenesis in DS group was 2 (median 2 with interquantile range 2-4). No statistically significant differences were found when considering gender in either group (P=0.85), neither in maxilla or mandible (P=0.89), nor in the right or left side (p=1).

The more frequently missing tooth observed in decreasing order was: maxillary lateral incisor, mandibular lateral incisor, mandibular second premolar, maxillary second premolar, mandibular central incisor, maxillary first molar and mandibular canine (Fig. 3).

Anomalies in size and shape of maxillary lateral incisor

Statistically significant differences were found for both size and shape anomalies (p<0.05). DS group showed 24.4% lateral anomalies whereas the control group showed 2.4% (Table 2). No statistically significant differences were found as far as gender nor right and left side.

Canine eruption path anomalies

The DS group showed 51.2% canine eruption path anomalies, as compared with 16.7% for the control group (p<0.05) (Table 2). Both canines were affected in 24.4% of DS subjects. No statistically significant differences were found for gender nor between the right and the left side. Regarding maxillary canines, 53.6% had already completely erupted in the control group, whereas only 8.5% had erupted in the DS group. Canine impaction was observed in 2.44% of the DS group and none in the control group. The control group showed transposition in 2.4%, as opposed to none in the DS group (Table 3).

Correlations

No significant correlations were found between size and shape of lateral incisor anomalies and canine path anomalies.

Discussion

Down Syndrome patients require special management. They need a careful and gradual approach, which takes time and patience. Therefore it is difficult to obtain reliable medical records for any research on their pathologies. In addition, not all the patients with DS allow neither a conventional dental and orthodontic exam nor an x-ray examination, consequently the sample size was relatively small. Selection of the sample

Distribution by sectors	DS (N=41)		Non DS (N=42)		Significance
Absolute Frequency	Right	Left	Right	Left	
Erupted	3	4	21	24	P<0.00001*
Sector I	23	23	15	13	
Sector II	7	8	5	5	Sector > II P=0.0008*
Sector III	8	2	0	0	
Sector IV	0	3	0	0	
impactation	0	1	1	0	
transposition	0	0	1	0	

TABLE 3 Distribution by sectors of upper canine eruption paths.

was carried out consecutively to prevent bias. The sample age was selected between 9 and 13 years of age to ensure a minimum age in the event of late dental formation, as in DS patients.

Agenesis

Thirty (30) out of 41 DS patients presented hypodontia (73%). As described in Table 4, this percentage is similar to that obtained in studies by Kumasaka et al. [1997], Bamba et al. [1994], Acerbi et al. [2001] and Shapira et al. [2000]. However, other studies, such as de Moraes et al. [2007], Cohen et al. [1965] and Sekerci et al. [2014], reported lower percentages. Given the delay in dental formation in DS patients, there is a risk of a lower detection of agenesis in samples of younger patients as well as in transversal studies based only on panoramic radiographs, due to a lack of medical information or study models. De Moraes et al. [2007] divided hypodontia in two types: proven and suspected, as they had no access to medical histories. Proven anodontia was considered when there was radiographic confirmation of presence of a primary tooth and absence of its permanent successor. Absence of a tooth without the presence of its primary predecessor was considered suspected anodontia. In cases of doubt the latter option was suspected. Fifty-three percent (53%) is obtained if the percentages of both types were added together. Sekerci et al. [2014] also found lower prevalence, which may be related to variation among different ethnic groups.

In agreement with other studies [Kumasaka et al., 1997; Bamba et al., 1994; Sekerci et al., 2014] no gender significant differences were observed. Some studies reported significant differences between upper and lower arches [Shapira et al., 2000; Russell and Kjaer, 1995; Reuland-Bosma, 2010; Sekerci et al. 2014]. In the present study, as in others, no significant differences between upper and lower arches, or between right and left side, were found [Kumasaka et al.,1 997; Bamba et al., 1994; Sekerci et al., 2014]. Distribution of developmentally absent teeth was similar to Jensen et al. [1973], de Moraes et al. [2007], Acerbi et al. [2001], Shapira et al. [2000] and Reuland-Bosma et al. [2010]. The maxillary lateral incisor was most commonly absent (31.8%). Other studies such as Kumasaka et al. [1997], Russell et al. [1995] showed that mandibular lateral incisor was most commonly absent. Error of identification may occur because of mesial drift of lateral mandibular incisors, implying dissimilar results between studies. To obviate this error by grouping together mandibular lateral and central incisors, the percentage changes to 30.5%, which remains lower than that for maxillary lateral incisors. The same percentage was found (17.6%) for mandibular lateral incisors and second premolar, followed by maxillary second premolars (14.1%), as in Jensen et al. [1973] and Acerbi et al. [2001]. Some other studies showed significantly more absent maxillary second premolars than mandibular second premolars [Kumasaka et al., 1997; Shapira et al., 2000; de Moraes et al., 2007; Sekerci et al. 2014]. The distribution pattern of developmentally absent teeth is similar to that of a normal population [Russell and Kjaer, 1995], with few exceptions such as maxillary first molars and mandibular canines. More absent teeth in incisor sectors than in premolar regions confirmed differences between DS patients and a normal population. Neither maxillary central incisors nor canines were absent when considering differences between upper and lower homologous teeth. These results may indicate that trisomy 21 plays a role in the induction of hypodontia. It has been reported that peripheral nervous system and cartilage alterations may explain the high frequency and distribution pattern of developmentally absent teeth in DS patients. Furthermore, the influence of congenital heart disease and hypothyroidism in tooth agenesis has recently been studied in DS patients [Reuland-Bosma et al., 2010].

Reference		Sample	Hypodontia	
Present study	DS	41	30	73%
	Control	42	3	7%
Cohen et al. 1965	DS	168	58	34.7%
	Control	---	---	---
Bamba et al. 1994	DS	128	92	72%
	Control	---	---	---
Russell et al. 1995	DS	100	8	81%
	Control	3325	258	7.8%
	Control	1530	126	8.2%
Kumasaka et al. 1997	DS	98	62	63%
	Control	150	26	17%
Shapira et al. 2000	DS	34	20	59%
	Control	---	---	---
Acerbi et al. 2001	DS	70	42	60%
	Control	---	---	---
De Moraes et al. 2007	DS	49	17	35%
	Control	---	---	---
Reuland-Bosma et al. 2010	DS	114	68	59.6%
	Control	---	---	---
Sekerci et al. 2014	DS control	216	57	26%
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TABLE 4 Differences obtained between DS and control groups in number and percentages of hypodontia.

Size and shape anomalies

Ten (10) out of 41 DS patients (24.4%) had size (microdontia) and shape anomalies of maxillary lateral incisors. These results differed significantly from the control group. Few studies have reported this kind of dental anomaly, specifically in maxillary lateral incisors [de Moraes et al., 2007; Cohen and Winner, 1965; Jensen et al., 1973]. These studies agree that prevalence is higher than in a normal population. Review articles [Desai, 1997] referred 35-55% microdontia of permanent dentition. The lower percentage found in the present study may be due to the fact that it was based only on dental history and not on clinical exploration or dental casts. Several studies [Shapira, 2000; Cohen and Winner, 1965 ; Jensen et al.,1973] have pointed out that a vascular alteration in the maxilla, which would affect odontoblast formation, would explain a higher presence of microdontia and shape anomalies in permanent dentition.

Upper canine eruption path anomalies

Twenty-one (21) out of 41 DS patients (51.2%) showed canine eruption path anomalies, which means that canine position was in sectors II, III and IV or was impacted according to the criteria of Lindauer et al. Canines were impacted in 2.4% of the cases, which means that root formation was completed but teeth were not erupted (not at occlusal plane level). This low prevalence cannot be explained by a genetic predisposition in the DS group. Shapira et al. [2000] found a higher percentage (15%), which may account for a lower mean age in the present study. Hence, with the knowledge that an existing delay in dental eruption in DS patients occurs, an increase in the mean age sample may show a higher percentage. Nevertheless, the present study also worked with a small sample consisting of 34 patients. No significant differences were observed as far as gender, nor side,as several studies reported.

Maxillary micrognathism, dental eruption delay and vascular alterations may account for a higher prevalence of canine eruption path anomalies [Shapira, 2000].

Regarding maxillary canine eruption pathology, evidence exists of the guidance theory of maxillary lateral incisor in the general population [Ericson and Kurol, 1988]. One of the aims of this paper was to demonstrate the existence of an association between size and form of maxillary lateral incisor and canine eruption path anomalies. However, no significant correlations have been found between these two variables. On the other hand, Shapira et al. [2000] found that 3 out of 5 canine impaction cases presented either agenesis or shape anomalies of maxillary lateral incisors. As previously mentioned, the present study used a small sample and therefore caution in inference is necessary. Reuland-Bosma et al. [2010], by contrast, found no association between canine impaction and maxillary lateral incisor agenesis.

Conclusions

This retrospective cross-sectional study based on panoramic radiographs and medical histories concludes the following:

1. Agenesis of permanent teeth in DS subjects occurs significantly more frequently than in non-syndromic subjects (73% vs. 7%).
2. No significant differences between upper and lower arch were found.
3. Maxillary lateral incisor was the tooh most frequently absent.
4. DS subjects had more significant size (microdontia) and shape (peg-shaped) anomalies of maxillary lateral incisors than the control group did (24.4% vs. 2.4%).
5. DS subjects showed more significant canine eruption path anomalies than the control group did (51.2% vs. 16.7%).
6. No significant gender differences were found for all variables studied.

Therefore, DS subjects would need periodical dental and orthodontic supervision with a panoramic follow-up at 6, 9, 12 and 15 years of age, so as to detect any anomaly and prevent or check for problems that may occur. We recommend a more in-depth assessment of all dentomaxilofacial anomalies in DS subjects and their multidisciplinary management with the aim of improving their dental health and their quality of life.

References

- Acerbi AG, de Freitas C, de Magalhäes MH. Prevalence of numeric anomalies in the permanent dentition of patients with Down Syndrome. Spec care Dentist 2001; 21:75-8.
- Bamba S, Maki Y, Ikeda M. Dental characteristics of Down syndrome patients. Part 2. Congenital absence of permanent teeth. J Jpn Soc Dent Handicap 1994;15:23-9.
- Cohen MM, Winner RA. Dental and facial characteristics in Down syndrome. J Dent Res 1965; 44:197-208.
- de Moraes ME, de Moraes LC, Dotto PP, dos Santos LR. Dental anomalies in patients with Down Syndrome. Braz Dent J 2007;18:346-50.
- Desai S. Down syndrome. A review of the literature. Oral Surg Oral Med Oral pathol Oral radiol Endod 1997; 84:279-85.
- Ericson S, Kurol J. Radiographic assessment of maxillary canine eruption in children with clinical signs of disturbances. Eur J Orthod 1986; 8:133-40.
- Ericson S, Kurol J. Early treatment of palatally erupting maxillary canines by extraction of the primary canines. Eur J Orhod 1988; 10:283-95.
- Jensen GM, Cleall JF, Yip AS. Dentoalveolar morphology and developmental changes in Down's syndrome (trisomy 21). Am J Orthod 1973; 64:607-18.
- Kumasaka S, Miyagi A, Sakai N, Sindo J, Kashima I. Oligodontia: a radiograpghic comparison of subjects with Down syndrome and normal subjects. Spec Care Dent 1997; 17:137-41.
- Lindauer SJ, Rubenstein LK, Hang WM, Andersen C, Isaacson RJ. Canine impactation identified early with panoramic radiographs. JADA 1992; 124:651-55.
- Menendez Nuñez M, Alarcon Perez JA, Gonzalez Rodriguez E. Estudio de la morfología cráneo-facial en el síndrome de Down. Ortod Esp 1992; 33:223-32.
- Musich DR. Orthodontic intervention and patients with Down Syndrome. Angle Orthod 2006; 76:735.
- Reuland-Bosma W, Reuland MC, Bronkhorst E, Phoa KH. Patterns of tooth agenesis in patients with Down syndrome in relation to hypothyroidism and congenital heart disease: an aid for treatment planning. Am J Orhtod Dentofacial Orthop 2010;137:584.e1-584e9.
- Russell BG, Kjaer I. Tooth agenesis in Down syndrome. Am J Med Genet 1995;55:466-71.
- Sekerci AE, Cantekin K, Aydinbelge M, Ucar FI. Prevalence of dental anomalies in permanent dentition of children with Down Syndrome. J Dent Child 2014 ;81:78-83.
- Shapira J, Chaushu S, Becker A. Prevalence of tooth transposition, third molar agenesis, and maxillary canine impactation in individuals with Down syndrome. Angle Orthod 2000;70:290-6.