

Three-dimensional hard tissue palatal size and shape in Down syndrome subjects

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SUMMARY The aim of the present study was to evaluate palatal morphology in Down syndrome (Ds) subjects, focusing on the effect of dental formula on the hard palate to assist clinicians when planning dental rehabilitation. Palatal landmarks were digitized with a three-dimensional (3D) computerized digitizer on the dental casts of 47 Ds subjects (23 dentate males, 9 edentulous males, and 15 dentate females) aged 20–45 years, 37 dentate reference individuals (20 males and 17 females) aged 30–39 years, and 14 edentulous reference males aged 55–72 years. The co-ordinates of the palatal landmarks were used to construct a mathematical equation of palatal shape, independent of dimensions. Palatal length, slope, width, and maximum palatal height in both the sagittal and frontal planes were measured.

In males, palatal length, width, and height were significantly influenced by both the syndrome and edentulism (analysis of variance, $P < 0.05$). The same measurements were significantly reduced in Ds compared with dentate females (t -test, $P < 0.05$). In the sagittal plane, Ds did not modify palatal shape; in the frontal plane, Ds individuals showed a higher palate. Overall, palatal shape was influenced by both Ds and edentulousness. Therefore, Ds seems to alter the normal palatal size and shape, although verification on larger samples is required. The findings of the present study may encourage more interdisciplinary dentofacial therapy in the dental and orthodontic care of Ds subjects.

Introduction

Down syndrome (Ds) is the most frequent chromosomal aberration in humans, with an incidence of 1.3 per 1000 live births, resulting from complete or partial trisomy of chromosome 21. In Italy, there are approximately 40 000 affected subjects, living in residential institutions and in the community; this last group is progressively increasing together with an improvement in their quality of life. Therefore, dental care for groups with special needs, such as Ds subjects, is a challenging problem in dentistry (Christensen, 2005; Glassman *et al.*, 2005).

Several maxillo-facial features have been described in subjects with Ds: a skeletal and dental Class III malocclusion, midfacial and mandibular hypoplasia, dental morphological and numerical anomalies, and eruption timing retardation of both the primary and the permanent dentitions (Peretz *et al.*, 1996, 1998; Uong *et al.*, 2001; Quintanilla *et al.*, 2002; Bagic and Verzak, 2003). Moreover, hypotonicity of the orofacial muscles is a common finding in these subjects, particularly in those who are institutionalized and who are likely to receive less stimuli from the environment (Glassman *et al.*, 2005). Hypotonia of the tongue muscles, together with their enlargement, often leads to tongue and mandibular protrusion and lastly to an open bite. In addition, a narrower, shorter, and relatively higher hard and soft palates have been described (Uong *et al.*, 2001; Carlstedt *et al.*, 2003).

Quantitative assessments of hard tissue palatal shape and dimensions in Ds subjects are still limited (Panchon-Ruiz

et al., 2000; Uong *et al.*, 2001; Skrinjaric *et al.*, 2004), and no data concerning Italian Ds subjects exist. Previous investigations of Ds individuals found a shorter hard palate length (Panchon-Ruiz *et al.*, 2000; Uong *et al.*, 2001), width, and height (Panchon-Ruiz *et al.*, 2000), and different palatal shape (Panchon-Ruiz *et al.*, 2000; Carlstedt *et al.*, 2003; Skrinjaric *et al.*, 2004). None of these studies reported on the dentition of the analysed Ds subjects, thus omitting the effect of the number of teeth on palatal morphology.

Computerized electromagnetic digitizers can provide three-dimensional (3D) co-ordinates of anthropometric landmarks and create databases for subsequent quantitative analyses (Ferrario *et al.*, 2001, 2003; Heiser *et al.*, 2004a,b; Dellavia *et al.*, 2006). These instruments appear sufficiently simple and fast for clinical application (Ferrario *et al.*, 2001, 2003; Heiser *et al.*, 2004a,b; Dellavia *et al.*, 2006).

In this investigation, the hard palate features of Italian institutionalized and non-institutionalized Ds subjects were analysed, using a computerized electromagnetic instrument which provides 3D metric co-ordinates of the chosen landmarks directly from dental casts (Ferrario *et al.*, 2001, 2003; Dellavia *et al.*, 2006). The data were used in mathematical and geometric models to distinguish size from palatal shape characteristics in comparison with reference population measurements. The effect of the number of the teeth on the Ds hard palate characteristics was also evaluated to provide information to assist in treatment planning.

Subjects and method

Subjects

Data from 47 Italian subjects with Ds (32 males, 15 females) aged 20–45 years [mean 32.27, standard deviation (SD) 12.54] were collected. Sixteen subjects lived in residential institutions (Sacra Famiglia, Cesano Boscone, Milano, Italy) and the other 31 were athletes attending the 2004 Special Olympics in Rome, Italy.

For the Ds males, the maxillary dentition ranged from almost a complete permanent dentition to complete edentulousness: nine individuals (six of them living in the residential institution) were completely edentulous and 23 (seven of them living in the residential institution) were partially dentate with an average of five anterior and six posterior maxillary teeth. Globally, the institutionalized males had fewer teeth than those living in the community.

In contrast, all Ds females had a minimum of two permanent maxillary molars per hemiarch. On average, Ds females living in residential institutions had two anterior and four posterior maxillary teeth, while those in the community had five anterior and seven posterior maxillary teeth. Palatal characteristics in Ds females were compared with those obtained from 'normal' females.

Reference data were collected from Italian healthy adults (20 males, 17 females) aged 30–39 years (mean 33.39, SD 2.31), with a sound full permanent dentition including the second molars, dental Class I, absence of crossbite, and no temporomandibular disorders or oral breathing. Additionally, measurements from edentulous reference palates were collected from 14 adult males aged 55–72 years (mean 64.67, SD 7.95). No subject had undergone craniofacial surgery.

To assess the influence of posterior teeth on palatal size and shape, palatal measurements were calculated separately for the partially dentate and for edentulous Ds males and compared with the dentate and edentulous reference males.

For all subjects with Ds, informed consent was obtained from the parents/legal guardians. All procedures were non-invasive and were approved by the local institutional review board.

Alginate maxillary impressions were taken for each subject and immediately poured in orthodontic white stone.

Collection of palatal landmarks

A specially devised protocol was used to examine both dentate and edentulous maxillary palates (Dellavia *et al.*, 2006). On each cast, the incisive papilla (IP) and most posterior limit of the right and left tuber maxillae (T_r , T_l) were identified. The midpoint (T_m) of the intertuber distance (T_r – T_l) was calculated and the IP– T_m line traced. This line was then divided into four equidistant segments and the relative transverse curves were traced to describe palatal

morphology. On IP– T_m , T_r – T_l , and the transverse curves, 12–14 almost equidistant points were then marked (Figure 1A; Dellavia *et al.*, 2006).

The 3D co-ordinates of IP, T_r , T_l , and 15–20 points on each of the four transverse and antero-posterior lines were obtained by a single operator (FO) with a computerized electromagnetic digitizer (3Draw, Polhemus Inc., Colchester, USA). The system has a resolution range of 0.0005 cm/cm and an accuracy of 0.025 cm, and supplies 'real' metric data independent of external reference systems.

To standardize and compare the palatal measurements for each cast, the plane described by IP, T_r , and T_l (X -axis: T_r – T_l line, right–left; Y -axis: antero–posterior; Z -axis: caudo–cranial) was set to horizontal. The following measurements were then computed for each palate:

Sagittal plane: (1) palatal length, horizontal projection of IP– T_m line (mm); (2) palatal slope, slope of the maximum palatal height versus the horizontal axis (degrees); and (3) maximum palatal height (mm).

Frontal plane: (1) palatal width at the tuber maxillae (T_r – T_l distance; mm) and (2) maximum palatal height (mm).

To describe palatal shape independent of palatal dimensions, all the co-ordinates in the sagittal plane were standardized as percentages of the horizontal projection of the IP– T_m distance (Y co-ordinate) and in the frontal plane as percentages of the intertuber distance T_r – T_l (X co-ordinate).

The palatal surface was then modelled by a four-degree polynomial $y = ax + bx^2 + cx^3 + dx^4$, both in the sagittal and in the frontal plane projections (Figure 1B): in the frontal plane, the origin of the axes was set at T_r (X -axis corresponding to T_r – T_l line; Y -axis to its vertical perpendicular) and in the sagittal plane, the origin of the axes was set at IP (X -axis corresponding to the horizontal projection of IP– T_m ; Y -axis to its vertical perpendicular; Ferrario *et al.*, 2003).

Error of method

The reliability of the measurements (intraoperator repeatability) was assessed by repeated tracings (landmark identification) and digitizations of the same casts. For each variable, the error of the method (error percentage) was calculated as the percentage ratio between the variance of the method error (squared Dahlberg's error) and the population variance of that measurement (squared SD). For landmark identification, the error percentage ranged between 0.5 and 1 per cent, and for landmark digitization between 0.38 and 0.63 per cent.

Statistical analysis

The mean and SDs were calculated for all measurements separately for each gender and group. Bivariate statistics

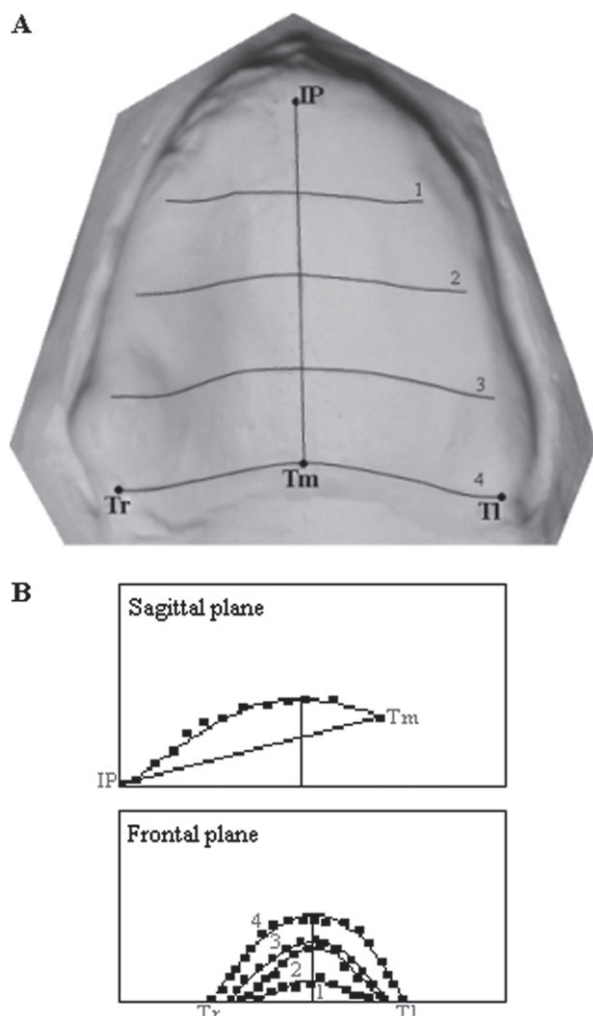


Figure 1 (A) Palatal landmarks marked on the stone cast of a completely edentulous Down syndrome male. IP, incisive papilla; T_r and T_l , right and left limits of tuber maxillae; and T_m , midpoint of the intertuber distance. (B) Palatal size and shape reconstruction using fourth-order polynomials of the same palate. Frontal view, IP– T_m line; lateral view, transverse curves.

with the rectangular components of the angles were used for slope values (Table 1).

To assess the normal distribution of all measurements within each group of subjects, the skewness and kurtosis were calculated. No large deviations from normality were found: the skewness ranged between +1.4 and –1.3, and kurtosis between 2.54 and 3.78.

For males, comparisons between groups were computed by a two-way factorial analysis of variance (factor 1: syndrome versus reference, factor 2: edentulism; syndrome versus reference $\times$ edentulism interaction). For females, palatal values computed in Ds subjects were compared with reference values by a two-tailed Student's *t*-test for independent samples. A level of significance of 5 per cent ($P \leq 0.05$) was used for all analyses.

Results

In males, both Ds and edentulism had a significant effect on several observed parameters (Table 2). Palatal length (IP– T_m) was shorter in Ds than in control males, and in dentate than in edentulous males ($P < 0.001$); the difference between dentate and edentulous individuals was larger in Ds males with a significant interaction between factors ($P = 0.003$).

Differences between Ds and reference palatal slope values were minimal (about 1 degree) in dentate subjects. In contrast, edentulous control individuals had a larger palatal slope than edentulous Ds males (interaction between factors, $P = 0.002$). The maximum palatal height in the sagittal plane showed significant differences between Ds and control subjects ($P < 0.001$) and between edentulous and dentate subjects ($P < 0.001$): the palate was higher in the controls than in the Ds males, and in dentate than in edentulous subjects; this difference was significantly larger for the Ds subjects (interaction between factors, $P < 0.05$).

Ds individuals had a smaller T_r – T_l distance than the reference subjects ($P < 0.001$). Also, edentulous subjects showed an increased palatal width ($P = 0.006$) in both groups (interaction between factors, $P = 0.03$).

Table 1 Palatal values in Down syndrome (Ds) and reference males.

Plane	Measurement	Reference ($n = 34$)				Ds ($n = 32$)			
		Dentate		Edentulous		Dentate		Edentulous	
		Mean	SD	Mean	SD	Mean	SD	Mean	SD
Sagittal	IP– T_m	50.23	2.69	45.66	2.97	43.43	4.44	37.26	5.75
	Slope	13.06	1.29	15.46	3.98	14.04	4.98	10.05	3.46
	Height	17.10	3.24	15.22	2.43	14.90	2.49	9.99	3.53
Frontal	T_r – T_l	45.59	3.46	48.11	3.00	35.59	7.42	38.01	4.65
	Height	12.38	4.50	13.28	3.05	12.79	3.98	7.44	2.57

Palatal measurements are in millimetres, except slope (degrees). Width: T_r – T_l ; length: IP– T_m ; SD, standard deviation.

In the frontal plane, all groups had almost the same mean values of maximum palatal height with great variability; only edentulous Ds individuals showed a reduction of approximately 5 mm in palatal height (interaction between factors, $P = 0.002$).

Table 3 summarizes the palatal measurements of Ds and reference females; data were compared with a Student's t -test ($P \leq 0.05$). On average, reference females had larger dimensions than Ds females. Statistical significance was found for palatal length ($P < 0.001$), maximum height in the sagittal plane ($P < 0.01$), and palatal width ($P < 0.001$). Palatal slope and maximum palatal height in the frontal plane were nearly superimposable for the two groups of females.

Globally, palatal dimensions were larger in dentate Ds males than in dentate Ds females.

Palatal shape was partially influenced by the presence of Ds; in Ds dentate subjects, the hard tissue palate was relatively higher than in the reference individuals in the frontal plane, obtained using the mathematical reconstruction with the four-order polynomials. The morphological modifications in the Ds subjects were similar for the whole palate; Figure 2A,B shows an example of the curves measured at the tuber maxillae. In edentulous males, a flattening of the palate was observed: Ds subjects had a smoother palatal curvature than reference subjects, particularly in the frontal plane (Figure 2C).

Table 2 Palatal data for males: two-way factorial analysis of variance.

Plane	Measurement	Down syndrome	Edentulism	Interaction
Sagittal	IP-T _m	0.001	0.001	0.003
	Slope	ns	ns	0.002
	Height	0.001	0.001	0.005
Frontal	T _r -T _l	0.001	0.006	0.030
	Height	ns	ns	0.002

Width: T_r-T_l; length: IP-T_m; 1, 62 degrees of freedom for all comparisons; ns, not significant ($P > 0.05$).

Table 3 Mean palatal measurements for females.

Plane	Measurement	Reference ($n = 17$)		Down syndrome ($n = 15$)		P
		Mean	SD	Mean	SD	
Sagittal	IP-T _m	46.29	3.08	40.77	3.74	0.001
	Slope	11.63	2.78	10.87	3.67	ns
	Height	13.76	1.86	11.00	3.34	0.01
Frontal	T _r -T _l	43.00	3.50	33.19	3.81	0.001
	Height	9.70	2.60	9.88	3.59	ns

Palatal measurements are in millimetres, except slope (degrees). Width: T_r-T_l; length: IP-T_m; SD, standard deviation. Comparison: Student's t -test for independent samples; ns, not significant ($P > 0.05$).

Discussion

The current study analysed the morphology of the hard tissue palate of Italian subjects with Ds using a computerized electromagnetic digitizer (Ferrario *et al.*, 2001, 2003; Dellavia *et al.*, 2006).

Globally, the average dimensions of the palate were significantly larger in reference males than in Ds subjects. In agreement with previous investigations (Panchon-Ruiz *et al.*, 2000; Uong *et al.*, 2001), the palate in the individuals with Ds was narrower, shorter, and lower in height. The measurements reported in previous studies are not directly comparable with the current findings because of the use of different instruments (callipers, moiré stripes, magnetic resonance) and protocols (reference landmarks). However, the present results seem to confirm a reduction in the palatal size of Ds subjects, as already assessed in other craniofacial structures (Desai, 1997; Farkas *et al.*, 2001, 2002; Bagic and Verzak, 2003; Sforza *et al.*, 2004; Ferrario *et al.*, 2005). Uong *et al.* (2001) found a mean reduction of 0.5 cm (0.86) in hard palatal length in Ds versus reference children and, as in the present study, Ds dentate adults (females plus males) had a diminished palatal width (mean 0.78), length (0.85), and maximum height (0.79). Panchon-Ruiz *et al.* (2000) found almost the same differences for all palatal dimensions measured at the first permanent molars (width 0.81, length 0.88, height 0.73) between young Ds adults of both genders and control subjects. Indeed, global diminished dimensions of the oral cavity with dental crowding and posterior crossbite have been described and mentioned as a cause of relative macroglossia in Ds subjects, together with an enlargement of the tongue and muscular hypotonia (Uong *et al.*, 2001; Carlstedt *et al.*, 2003).

The current study analysed the palatal morphology of Italian Ds individuals, while previous investigations have analysed North American (Uong *et al.*, 2001), Swedish (Carlstedt *et al.*, 2003), Spanish (Panchon-Ruiz *et al.*, 2000), and Croatian (Skrinjaric *et al.*, 2004) Ds subjects. In these different groups, all palatal dimensions were diminished.

Overall, all palatal dimensions were somewhat larger in Ds males than in Ds females with molar teeth, in agreement with the literature (Panchon-Ruiz *et al.*, 2000; Skrinjaric *et al.*, 2004). The sexual dimorphism observed in the reference subjects in a previous investigation (Ferrario *et al.*, 2001) was also confirmed in the Ds subjects.

The protocol used in the current investigation was devised specifically to examine palatal morphology independent of dental landmarks, and was therefore suitable for both dentate and edentulous maxillary arches. Indeed, Ds subjects often lack molar teeth, while it is extremely rare to find complete edentulousness in non-syndromic young adults (Peretz *et al.*, 1996; Desai, 1997; Quintanilla *et al.*, 2002). Only limited data on palatal size and shape in edentulous reference subjects are available in the literature, but these are from an elderly population (Klemetti *et al.*, 1996). Also

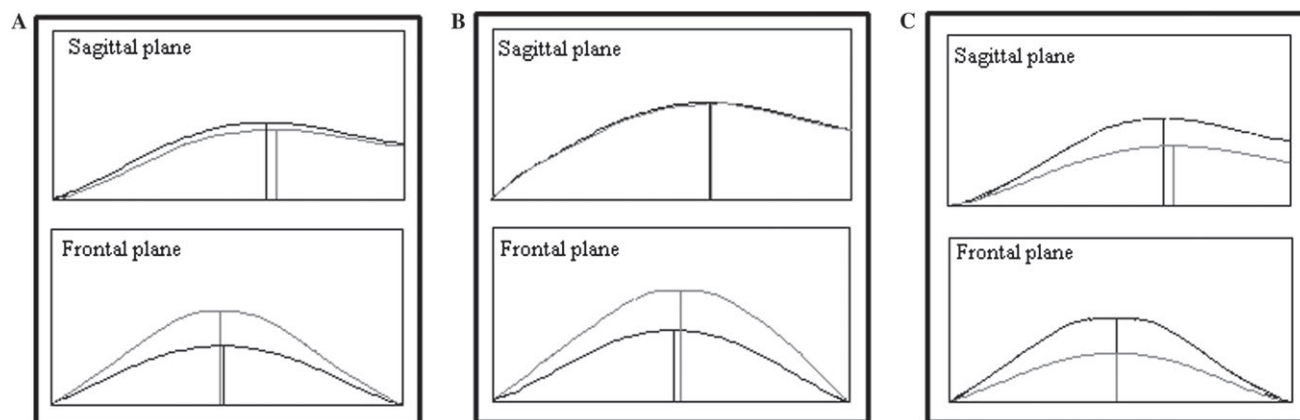


Figure 2 Mean palatal shape (size-independent) in females (A), dentate males (B), and edentulous males (C). Down syndrome (black lines) and reference individuals (grey lines). Frontal and lateral (curve between T_r and T_l) views. X-axis unit = percentage of intertuber distance; Y-axis unit = percentage of IP- T_m distance.

in the present study, the palatal casts of the edentulous reference individuals were obtained from older subjects, thus introducing a possible confounding effect of age.

The present group of Ds subjects included a wide range of dentitions, thus permitting evaluation of the relative contribution of posterior teeth and syndrome to hard palate morphology. To assess the role of molar teeth on palatal features, Ds males were divided into two groups: one completely edentulous and the other having a dentition comparable with healthy subjects of the same age. Most measurements were smaller in partially dentate Ds compared with the reference dentate individuals and in edentulous Ds compared with the edentulous control individuals: trisomy of chromosome 21 seemed to affect palatal dimensions. After maxillary tooth extraction/loss, the residual alveolar ridge undergoes rapid atrophy, thus causing palatal vault remodelling; the IP gradually moves in an anterior direction together with a flattening of the palatal surface (Klemetti *et al.*, 1996; Heiser *et al.*, 2004a,b). Additionally, the majority of the Ds subjects living in residential institutions analysed in the current investigation did not wear any prosthesis and showed greater atrophy of the upper alveolar crest.

The number of missing permanent teeth of institutionalized Ds patients compared with non-institutionalized Ds patients documented in this study should encourage more interdisciplinary dentofacial therapy when these patients are younger. Early therapy that provides Ds patients with arch expansion, tooth preservation, correction of impacted canines, and replacement of congenitally absent teeth may permit a significantly improved quality of life for ageing Ds patients who seem to have a longer lifespan (Desai, 1997).

Additionally, the present study analysed palatal shape alterations using fourth-degree polynomials (Ferrario *et al.*, 2001, 2003; Dellavia *et al.*, 2006): in a lateral view, within each gender the curves of Ds and reference dentate subjects were nearly superimposable; in a frontal view, Ds individuals showed a higher palate than control subjects (Figure 2A,B).

Carlstedt *et al.* (2003) found wide variability in the formal characteristics of the hard palate analysed from a qualitative point of view (flat, staired, round, high, pointed, and narrow) with a majority of staired palates (65 per cent) in Ds children. Skrinjaric *et al.* (2004) also observed a higher frequency of staired palates in young Ds patients (3–14 years), but this finding significantly decreased with age.

Considering the influence of teeth on the palate of Ds subjects, shape modifications were larger in edentulous subjects. In both frontal and lateral views, Ds individuals without molar teeth had smoother palatal curves than reference edentulous males with a more relevant flattening of palatal curvature and a lateral displacement of the tuberosity landmarks (Figure 2C).

The present quantitative data on palatal size and shape could represent reference values (Glassman *et al.*, 2005). For example, the high vault and narrow arch documented in this study, through digitized models, should encourage orthodontists to further evaluate the transverse skeletal basal bone deficiency in patients with Ds through the use of antero-posterior cephalograms.

Currently, more children with Ds are not being institutionalized and as a result, the dental community may have more opportunity to provide care to preserve teeth in this group of patients who frequently have several congenitally missing teeth (Christensen, 2005). Tooth preservation would reduce the accelerated alveolar atrophy which makes prosthetic replacement more challenging (Glassman *et al.*, 2005).

Palatal growth in Ds subjects should also be investigated to help in treatment planning. Knowledge of the alterations in skeletal development of the maxillary arch and palate at different ages is an essential parameter to decide the correct timing of functional rehabilitation. Therefore, the effect of the number of teeth in Ds females and age-related variations need to be evaluated in the future.

Conclusions

Ds seems to alter the normal size of the hard tissue palate with a reduction of all parameters. Shape modification consists of a relatively higher palatal vault. However, further analyses of larger groups are necessary to obtain a better insight into palatal morphology in Italian Ds subjects, thus offering useful information to the clinician.

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