Original Article

Longitudinal maxillary growth in Down syndrome patients

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ABSTRACT

Objective: To assess maxillary growth in a sample of patients diagnosed with Down syndrome (DS).

Materials and Methods: The sample comprised 47 subjects (25 boys, 22 girls) with DS. All patients had at least two radiographs that showed the cranial base. To obtain comparisons among age groups, the sample was divided into three groups: prepubescent (8–11 years old), pubescent (12–14 years old), and postpubescent (15–18 years old). A control group included 38 subjects without DS (22 boys, 16 girls) who were part of a longitudinal growth sample. Computerized cephalometric analysis was performed on all subjects, and cephalometric superimpositions were made. Two-way analysis of variance (ANOVA) was used to study the overall changes between groups. In addition, one-way ANOVA and the Duncan multiple-range test were used to analyze possible differences in the age groups.

Results: Sagittal maxillary growth in DS patients was constant from the age of 8 to 18 years; there was an average increase of 0.12 mm/year, measured at the level of point A. In the vertical plane it grows at an average rate of 0.62 mm/year and 0.70 mm/year, measured at the level of the ANS and PNS, respectively.

Conclusions: The maxilla in the DS group shows hypoplasia in the vertical plane and the sagittal plane, and there was a mean deficit of almost 10 mm in the latter. (*Angle Orthod.* 2011;81:253–259.)

KEY WORDS: Down syndrome; Maxillary; Growth; Cephalometry

INTRODUCTION

Down syndrome (DS), described by Down in 1866, was found to be an autosomal chromosome abnormality related to the trisomy of the 21st pair by Jerome Lejeune in 1959.¹ With an incidence of 1 per 1250

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births,² it is the most frequent chromosome alteration, and the average life span of persons with DS is around 57 years.

Many studies report the prevalence of malocclusions that exists among people with Down syndrome.³⁻⁶ More specifically, there is a greater frequency of anterior and posterior crossbites, Class III molar relationships, and anterior open bite in persons with DS than in persons with other types of mental handicaps or in the general population.

Since 1932, when Broadbent⁷ applied teleradiographic techniques to orthodontics, cephalometric analysis has become the main diagnostic tool for orthodontists and an accurate method to study and assess the changes that take place due to growth and/ or treatment in the different craniofacial structures.

However, despite the numerous studies on craniofacial growth carried out on the general population, there has been no comparable study carried out on people with DS. Cephalometric analyses of patients with DS are rare and are carried out on cross-sectional samples, often over a wide range of ages, but none of them offer a dynamic vision of craniofacial growth as a whole. Therefore, the objective for this study is to describe the changes in the bone of the craniofacial complex that take place in the sagittal and the vertical planes due to spontaneous growth in people with DS. The present study is limited to the maxilla and compares the results with a longitudinal sample of facially normal persons.

MATERIALS AND METHODS

The experimental group included 47 subjects (25 boys and 22 girls). The selection criteria for the sample were that they had been diagnosed with DS and were still growing at the time of the first radiograph. At least two radiographs were taken of each patient. The average age of the group in the initial radiograph was 11 years, 8 months (ranging from 7 years, 7 months to 16 years, 4 months). Some patients older than 16 years were included to try to identify late growth spurts that have been found in these patients.⁸ The average age at the last radiograph was 14 years, 1 month (ranging from 8 years, 7 months to 18 years, 6 months). To draw comparisons between the different stages of growth, the sample was divided into three age groups: prepubescent (8-11 years old), pubescent (12-14 years old), and postpubescent (15-18 years old). This classification was made according to other authors who believe the pubescent period in DS occurs similarly to that of the general population, although with a lower rate of growth.9,10

The control group included 38 subjects who satisfied the following criteria: still growing at the time of the first radiograph, no apparent craniofacial deformities, no history of craniofacial trauma or congenital anomalies, occlusal stability with clear intercuspation, no extractions of permanent teeth, no dental anomalies, and no maxillofacial surgery or surgical treatment. They were also born in Spain or a direct descendent of Spaniards. This group included 16 girls and 22 boys. The average age at the first radiograph was 10 years, 4 months, for the girls and 10 years, 1 month, for the boys. At the last radiograph, the ages were 13 years, 6 months, for the girls and 13 years, 8 months, for the boys.

At least one lateral cranial radiograph was taken of each patient annually for as long as he or she participated in the study. All the radiographs were done using the same machine, with a magnification index of 1:1.10 (10%).

In all lateral cranium radiographs, a cephalometry was traced with a computerized analysis (Nemotec Dental System, Nemoceph Studio, Madrid, Spain) by marking the following cephalometric landmarks (Figure 1): S (sella), N (nasion), Ba (basion), ANS (anterior nasal spine), PNS (posterior nasal spine), point A, Po (porion), Or (suborbital), CC (pterygomaxillare), and



Figure 1. Cephalometry traced with a computerized analysis.

Co (condylion). Table 1 shows the cephalometric parameters traced using these landmarks.

To determine the growth value, we used the superimposition¹¹ of the initial and final lines drawn with the Ba-N plane coinciding at point N. Changes in point A were measured against the Frankfort horizontal plane as a horizontal reference, and the changes in the ANS and PNS were measured against the vertical pterygoid as a vertical plane of reference. A positive value was given to point A when its displacement was forward. A positive value was given to the vertical changes when the displacement of the nasal spines was downward.

We also recorded the rotational changes of the maxilla using the palatal plane (ANS-PNS). The rotation was considered positive when the final position of the palatal plane had undergone a counterclockwise turn with respect to the initial position (Figure 2).

All cephalometric analyses were made by two researchers (JL and CL) from a project of the general growth study in the orthodontics masters program at the Universidad Complutense de Madrid. These researchers calibrate their instruments annually to prevent errors in the cephalometric measurements. After they measured the lines, they compared the findings and obtained these possibilities: (1) concordance type I, absolute coincidence of the 2 lines; (2) concordance type II, some difference in the parameters of the 2 lines—Co-A (effective maxillary length), <1 mm; N-Cf-A angle (maxillary height), <2°; N-ANS (height of midfacial area), <1 mm; SNA angle, <2°; Frankfort/N-A plane angle (maxillary depth), <2°;

Table 1.	Cephalometric	Parameters	Used in	this	Study
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Cephalometric Parameters	Definition				
SNA	Sella-nasion-point A angle				
Maxillary depth	Angle between Frankfort and nasion-point A				
Distance point A	Distance from point A McNamara's line to Frankfort from N				
Effective maxillary length (Co-A)	Distance condylion-point A				
Palatal plane length	Distance anterior nasal spine to posterior nasal spine (ANS-PNS)				
Maxillary height	Angle N-Cf-point A				
Height of the midfacial area (N-ANS)	Distance nasion to ANS				

distance of point A to McNamara's line from N, $<\!1$ mm; and (3) concordance type III, a greater difference than previously described.

In concordance type II, the arithmetic mean between the two values is established for the parameter that does not coincide. When the difference is greater (concordance type III), the lines are drawn, and the measurements are made again, bearing in mind the three types of concordance. Standard error was determined using the Dahlberg formula $(SE = \sqrt{d^2/2n})^{12}$ and the systematic error was found using the Student's *t*-test at P < .05.¹³ A descriptive statistical analysis was carried out to evaluate the data obtained that included the arithmetic mean, percentage, and range (maximum and minimum values) of each variable for each group (DS and control) according to sex and age. Then an analytical or inferential statistical analysis was done in which the differences between the two groups were analyzed by using the Student's t-test for independent samples to establish comparisons between age groups. To study the evolution of each variable over time and to establish comparisons in the behavior of one particular variable in each group, we used the twoway analysis of variance (ANOVA) with interaction.

In each group and to compare the different age groups, we applied one-way ANOVA followed by the



Figure 2. Rotational changes of the maxilla and cephalometric superimpositions.

Duncan multiple-range test as an a posteriori test with a 0.05 confidence level. To determine the differences between the sexes, we also used the Student's *t*-test.

RESULTS

Table 2 gives average values, standard deviations, and the statistical significance of the values obtained from the cephalometric superimposition method. It shows that there are significant differences between the groups for the total sample and for the three age groups for the variables that in some way measure maxillary size on the sagittal plane (Co-A and ANS-PNS) and vertically (N-Cf-A and N-ANS angle).

In Table 3, the only statistically significant differences appeared in the DS group: Co-A (effective maxillary length), ANS-PNS (palatal plane length), and N-ANS (height of the midfacial area). All three of these variables were greater in boys.

Table 4 shows the growth pattern and indicates that the way in which the maxilla grows in patients with DS is very similar to that of people who do not have DS.

The main finding of the study was that there was a deficit of almost 10 mm in both sagittal measurements (Co-A and ANS-PNS). In addition we also found difference of 2.5° and almost 8 mm for Ricketts' maxillary height and N-ANS, respectively, and these values were always smaller in people with DS (Table 2). All these measurements have been analyzed in greater detail in the discussion section.

DISCUSSION

Sagittal Maxillary Growth

The three parameters used to situate the maxilla in the front and back with respect to the cranial base (SNA, maxillary depth, and distance to McNamara's point A) are within the normal parameters and did not show significant differences compared to the control group in the value of the measurement or in how they evolved over time (Tables 2 and 4). In other words, the placement of the maxilla in relation to the cranial base between the ages of 8 and 18 years is similar to that of the general population. This finding agrees with other cephalometric studies, such as those done by Fischer-Brandies and colleagues,^{8,14} in which they observed a

Table 2. Overall Results of the Sample According to Age Groups^a

Maxillary Parameters		Total (8–18 y) Prepubescent (8-		t (8–11 y)	-11 y) Pubescent (12-14 y)			Postpubescent (15–18 y					
Variable	Group	Mean	SD	Р	Mean	SD	Р	Mean	SD	Р	Mean	SD	Р
Superposition													
A (mm/y)	DS	0.12	0.24	NS	0.14	0.20	NS	0.12	0.14	NS	0.11	0.32	NS
	Control	0.20	0.29		0.22	0.31		0.19	0.28		0.11	0.21	
ANS (mm/y)	DS	0.62	0.76	NS	1.15	0.93	NS	0.72	0.65	NS	0.15	0.37	<i>P</i> < .05
	Control	0.98	0.65		1.05	0.47		0.97	0.85		0.48	0.25	
PNS (mm/y)	DS	0.70	0.77	NS	1.05	0.78	NS	0.92	0.85	NS	0.25	0.38	NS
	Control	1.02	0.58		1.09	0.49		1.03	0.68		0.49	0.27	
Palatal plane (degree/y)	DS	-0.04	0.74	NS	0.05	0.83	NS	-0.24	0.62	P < .05	0.08	0.75	NS
	Control	0.08	0.68		0.08	0.75		0.11	0.63		-0.09	0.10	
Cephalometry													
SNA (degree)	DS	78.85	3.44	NS	77.16	3.18	<i>P</i> < .01	79.74	3.57	NS	79.24	3.15	NS
	Control	79.27	3.19		78.74	2.67		79.72	3.47		79.68	3.86	
Maxillary depth (degree)	DS	91.49	3.21	NS	90.42	3.67	NS	91.72	3.68	NS	91.99	2.29	NS
	Control	90.55	2.69		90.19	0.69		90.82	2.65		90.94	2.78	
Distance A (mm) (mm)	DS	1.31	2.86	NS	0.28	3.14	NS	1.51	3.27	NS	1.82	2.14	NS
	Control	0.54	2.81		0.20	2.74		0.88	2.83		0.62	3.03	
Effective maxillary length	DS	78.89	5.79	<i>P</i> < .001	74.21	5.04	<i>P</i> < .001	79.75	5.09	<i>P</i> < .001	81.22	5.01	<i>P</i> < .001
(mm)	Control	88.57	4.65		86.30	4.22		90.02	4.18		92.15	3.54	
Palatal plane length (mm)	DS	43.33	3.37	<i>P</i> < .001	40.91	2.81	<i>P</i> < .001	43.61	3.56	<i>P</i> < .001	44.65	2.69	<i>P</i> < .001
	Control	53.23	3.46		51.48	2.88		54.42	3.15		55.71	3.42	
Maxillary height (degree)	DS	56.23	4.12	<i>P</i> < .001	56.04	4.66	<i>P</i> < .001	55.72	3.55	<i>P</i> < .001	56.72	4.18	<i>P</i> < .001
	Control	58.88	3.01		58.41	3.01		59.13	2.95		59.85	3.02	
N-ANS (mm)	DS	46.52	3.90	<i>P</i> < .001	44.13	4.29	<i>P</i> < .001	46.28	3.18	<i>P</i> < .001	48.23	3.27	<i>P</i> < .001
	Control	54.30	3.53		52.52	3.33		55.50	3.04		56.86	2.52	

^a NS indicates not significant; DS, Down syndrome; ANS, anterior nasal spine; PNS, posterior nasal spine; n, nasion; s, sella.

Maxilla		Females		Ма	les	
Variable	Group	Mean	SD	Mean	SD	Р
Superposition						
A (mm/yr)	D	0.10	0.18	0.14	0.28	NS
	С	0.29	0.37	0.13	0.18	<i>P</i> < .01
ANS (mm/yr)	D	0.64	0.79	0.60	0.75	NS
	С	0.71	0.47	1.21	0.69	<i>P</i> < .001
PNS (mm/yr)	D	0.73	0.75	0.68	0.79	NS
	С	0.84	0.61	1.18	0.51	<i>P</i> < .01
PP (degree/yr)	D	-0.13	0.78	0.02	0.70	NS
	С	0.07	0.69	0.08	0.67	NS
Cephalometry						
SNA (degree)	D	78.30	2.81	79.28	3.82	NS
	С	78.44	3.53	79.92	2.75	NS
Max depth	D	91.69	2.78	91.32	3.53	NS
(degree)	С	90.69	3.05	90.44	2.37	NS
Dist A (mm)	D	1.45	2.24	1.20	3.29	NS
	С	0.77	3.18	0.36	2.48	NS
Ef Max length	D	75.08	3.49	81.93	5.46	<i>P</i> < .001
(mm)	С	87.06	4.34	89.76	4.56	<i>P</i> < .001
PP length	D	41.17	2.03	45.05	3.23	<i>P</i> < .001
(mm)	С	52.53	2.66	53.79	3.91	P < .05
Max height	D	56.39	2.28	56.10	5.15	NS
(degree)	С	59.45	2.81	58.44	3.09	P < .05
N-ANS (mm)	D	44.90	2.83	47.81	4.15	<i>P</i> < .001
	С	54.69	3.41	53.98	3.61	NS

Table 3. Differences According to Sex

^a NS indicates not significant; DS, Down syndrome; ANS, anterior nasal spine; PNS, posterior nasal spine; n, nasion; s, sella.

similar SNA angle in patients with DS and chromosomally normal subjects. They all found that this angle remained constant at all ages. We also found a slight increase in maxillary depth and in the distance to McNamara's point A in the group with DS, but in line with this hypothesis, we consider this characteristic of the fact that point N is farther back because the anterior cranial base in patients with DS is typically shorter than that of the rest of the population, as is attested to by many authors.¹⁵

To study sagittal maxillary growth independently of the anterior cranial base, the cephalometric superimpositions were done assessing the evolution of the point A projection over the Frankfort plane (Figure 2). The results obtained from this procedure were not significantly different in the two groups. We found yearly increases very close to 0, especially from the age of 15 years, when the three parameters used to study the maxilla in the sagittal plane (SNA, maxillary depth, and distance to McNamara's point A) become more stable (Table 2). Braun and colleagues¹⁶ showed that sagittal growth in the general population tended to be smaller after the age of 14 years and that it stopped completely at the age of 16 years.

All of the aforementioned allows us to affirm that sagittal maxillary growth is closely related to cranial base growth in patients with DS just as in the control

Table 4.	Evolution	of Maxillary	Parameters	Over	Time
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Variable	Group	Evolution ANOVA (2×2)	Prepubescent (8–11 y)	Pubescent (12–14 y)	Postpubescent (15–18 y)
Superposition	· · ·	/	,		
A	DS	NS	NS	NS	NS
	Control		NS	NS	NS
ANS	DS	NS	P < .05	P < .05	P < .05
	Control		NS	NS	NS
PNS	DS	NS	NS	NS	P < .05
	Control		NS	NS	P < .05
Palatal planee	DS	NS	NS	NS	NS
	Control		NS	NS	NS
Cephalometry					
SNA	DS	NS	P < .05	NS	NS
	Control		NS	NS	NS
Maxillary depth	DS	NS	P < .05	NS	NS
	Control		NS	NS	NS
Distance A	DS	NS	P < .05	NS	NS
	Control		NS	NS	NS
Effective maxillary length	DS	NS	P < .05	NS	NS
, ,	Control		P < .05	P < .05	P < .05
Palatal plane length	DS	NS	P < .05	NS	NS
	Control		P < .05	NS	NS
Maxillary height	DS	NS	NS	NS	NS
	Control		NS	NS	NS
N-ANS	DS	NS	P < .05	P < .05	P < .05
	Control		P < .05	NS	NS

^a NS indicates not significant; DS, Down syndrome; ANOVA, analysis of variance; ANS, anterior nasal spine; PNS, posterior nasal spine; n, nasion; s, sella.

subjects. Both structures move forward coordinately, as was also observed in longitudinal studies of chromosomally normal patients carried out by Walker and Kowalski,¹⁷ Bishara,¹⁸ and Bishara and Jakobsen.¹⁹

Vertical Maxillary Growth

We studied vertical maxillary growth with reference to maxillary height and height of the midfacial area using the projections of the anterior and posterior nasal spines over the pterygoid vertical.

The maxillary height (N-CF-A angle) and the height of the midfacial area (N-ANS) show that the vertical dimension of the nasomaxillary complex is significantly smaller in subjects with DS (Table 2), which establishes the existence of hypoplasia in the vertical plane. Farkas and colleagues²⁰ also found these results.

However, the descent of the posterior and anterior nasal spines, which takes place during growth in patients with DS was similar to that in the control group until the age of 14 years. It is in the postpubescent stage that the growth rate falls in the group with DS, especially for the ANS. In other words, until that time the maxilla grows vertically at the same rate and with the same intensity in both groups. Therefore, the vertical deficit of the maxilla in the group with DS must be present in earlier ages than those included in this study. The pronounced descent in the postpubescent stage indicates a tendency for vertical growth to stop at an earlier age in people with DS than in the general population (Tables 2 and 4 and Figure 2).

The ANS and PNS in patients with DS descend at practically the same rate, at 0.62 and 0.70 mm/y, respectively. This means the maxilla does not undergo any rotation in its descent during facial development. This interpretation agrees with the analysis of the palatal plane in the superimpositions, which shows a negligibly small oscillation in both groups (consistently close to zero $[\pm 0.2^{\circ}]$; Tables 2 and 4).

Our results square with studies done by Fischer-Brandies and colleagues,^{8,14} who observed that the maxilla descends vertically during growth, but they found no rotation in persons with DS.

Regarding differences between the sexes, even though the girls revealed shorter height of the midfacial area than the boys, there was not significantly different growth in function of sex in persons with DS (Table 3).

Maxillary Size

Both Co-A (effective maxillary length) and ANS-PNS (palatal plane length) were significantly diminished in patients with DS for the duration of our study. Maxillary size, as shown by these two variables, increases, as with that of the control group, but with an important difference in amount of about 10 mm (Table 2).

These observations coincide with classic studies by several authors, including Redman and colleagues²¹ and Westerman and colleagues,²² in which they describe a three-dimensional small palate as characterized in DS using plaster models. Using lateral radiographs, Baer and colleagues¹⁵ reported minimal hard palate growth between the ages of 6 and 13 years in these individuals. In cephalometric studies, Fischer-Brandies⁸ confirmed maxillary hypoplasia in the sagittal and vertical planes. Farkas and colleagues^{20,23} and Allanson and colleagues²⁴ also reached the conclusion that patients with DS have a severe decrease in the height and depth of the midfacial area.

If the maxilla grows in accordance with the cranial base, and hypoplasia is present in both structures, the SNA angle and the rest of the variables that relate the maxilla to the cranial base (facial depth and distance from McNamara's point A) remain unaltered within the cephalometric norm.

However, in spite of hypoplasia, the evolution over time of effective maxillary length and palatal plane length was similar in both groups, with parallel lines on the charts (Table 4 and Figure 2), which means that patients with DS start from a deficient initial situation, but the relationship stays quite stable over time. That is, both groups had the same kind of growth in the period of observation. In other words, these individuals present, as we have seen, a hypoplastic maxilla in the vertical plane and even more so in the sagittal plane, but it grows at the same rate as in the general population.

According to sex, independently of hypoplasia, boys with DS had bigger maxillae than girls (Table 3), thus showing that sexual dimorphism exists in relation to maxillary size. This difference was also present in the control group and agrees with the results obtained by Ursi and and colleagues²⁵ in their study on craniofacial growth done with chromosomally normal individuals.

CONCLUSIONS

- The relationship of the maxilla with the cranial base in DS is totally normal and grows in the sagittal plane in coordination with the cranial base.
- People with DS have maxillary hypoplasia, especially in the horizontal plane, but also in the vertical plane.
- The developmental deficiency in the midfacial area in patients with DS is established before the ages considered in our study and persists throughout the growth period.
- Between 8 and 18 years of age, maxillary growth in patients with DS is similar to that in the general population, but it starts with a less developed bony structure.

- Maxillary growth tends to cease at an earlier age in persons with DS compared to the general population.
- There is some sexual dimorphism in patients with DS because the measurements in males are longer than in females, but it is comparable to that found in the general population.

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