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Rapid maxillary expansion and nasal patency in children with Down syndrome*

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SUMMARY

Down syndrome (DS) is the most common aneuploid disorder at birth. The life expectancy of persons with DS has improved over the last forty years and is now at about sixty years. Phenotypic characteristics include general hypotonia, maxillary hypoplasia with a small oral cavity and a somewhat larger appearing tongue, frequent constricted maxillary arch, nasal obstruction and others. This prospective study assesses the effects of rapid maxillary expansion (RME) on nasal patency of children with DS, using acoustic rhinometry (AR). Twenty four children with DS, aged 5 to 12 years, had been randomly allocated to the RME and control groups. AR was performed to these individuals prior to expansion, approximately one month after, post maximal expansion, and after a 5 months period of retention. The data between the two groups were compared. Rapid maxillary expansion produced a significant augmentation of nasal volume in children who had been treated ($p<0.05$) compared to the control group; these results were stable through the period of retention.

Key words: acoustic rhinometry, Down syndrome, maxillary expansion, paediatric

INTRODUCTION

Down syndrome (DS) is the most common aneuploid disorder in live born infants with a prevalence of 1/770 live births[1]. The life expectancy for individuals with DS has substantially increased over the last four decades. Now they are often living until the 6th decade and this is contributing to an increased prevalence[2]. This has several implications for service providers, particularly for special support in health, education and social services in order to attain a better quality of life for these individuals. Phenotypic characteristics include mental retardation, general hypotonia, maxillary hypoplasia with a larger appearing tongue, often obesity that may result in specific otolaryngologic symptoms such as upper respiratory obstruction, sleep apnea and others[3].

Rapid maxillary expansion (RME) is an orthodontic procedure used to correct the narrow transverse diameter leading to a widening of the arch perimeter that will provide more space for alignment of crowded teeth and also permits the correction of crossbite. Although the major effect of treatment is noticed clinically in the area of dentition, transverse enlargement of the maxillary bone may be considered an additional benefit including the nasal width[4]. Usually these results are directly related to changes in nasal airway flow that will improve nasal ventilation[5, 6, 7, 8].

Acoustic rhinometry (AR), introduced by Hilberg et al. in 1989, is based on the reflection of an acoustic signal introduced into the nasal cavity. It can be used to evaluate the cross sectional area of the nasal cavity and enables the calculation of the nasal volume[9]. AR is a reliable method to measure the dimensional changes of the nasal cavity before and after a given treatment and thus will provide valuable information about skeletal variation of the nose[10].

This study analyses the effect of RME on nasal patency in a group of children with DS. A multicentre research ethics committee approved this study. To the best of our knowledge this is the first such investigation performed in children with DS.

MATERIALS AND METHODS

Patients

The recruitment of children with Down syndrome was done by mailing detailed study information to the main organizations working with this population in Portugal. Two centers

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were set up, one in the south, Lisboa, and the other in the north, Porto. An otolaryngologist and a paediatric dentist were examining the children. A questionnaire was completed for each of the 106 children in the study recording otolaryngological symptoms, particular paying attention to upper airway obstruction. The paediatric orthodontist studied maxillary compression and dental cross bite.

The inclusion criteria were: (i) cytogenetic diagnosis of trisomy 21, (ii) age between 4 and 12 years, (iii) persistent nasal obstruction and/or repeated upper respiratory infections of three episodes over 6 months or four episodes in a year, (iv) presence of lateral crossbite and/or evidence of maxillary compression, (v) adequate cooperation, (vi) availability of frequent follow-up examinations, and (vii) informed consent from the children's legal representatives.

Acoustic rhinometry

From the total group of children with Down syndrome, 26 were selected and divided into subgroups according to age (4 to 6, 7 to 10, and 10 to 13 years) to avoid possible bias on the final results. Children from each of these sub-groups were randomly assigned to two groups: RME and control. They underwent acoustic rhinometry which was always performed by the same examiner. The instrument used for these examinations was an Eccovision Acoustic Rhinometer with a software version 3.6 supplied by Hood Laboratories. The procedure was standardized by measuring at baseline and approximately ten minutes after nasal decongestion using a mixture of 0.25% oxymetazoline. Cast polyurethane nose pieces were used to fit the size and shape of the nostrils of the child. The probe was aligned near the midline about 45° from the vertical. No sealant was used. Children were asked not to move and to hold their breath. The mean curve of 10 measurements +/- one standard deviation was obtained. As main parameter the nasal volume at 0 to 4 cm was measured, because it represents an integration of several cross-sectional areas [10].

Maxillary expansion

An intraoral device was applied to 13 children in the RME group. Rapid maxillary expansion was accomplished with an individually designed appliance that was easy to clean[11]. It was usually fixed with orthodontic banding to the posterior teeth, first molars or the corresponding primary teeth. The two maxillary bones were separated at the midline suture by mean of a screw mechanism located in the midline of the appliance (Figure 1). Activation rates of the order of 0.3 to 0.5 mm per day permitted painless separation at the mid-palatine suture in the children.

Clinical examination and AR were performed in the RME group before the intraoral application of the device at time 0 (T0), approximately one month after the start of the rapid total expansion at time 1 (T1) and after the retention period of 5 months at time 2 (T2). The same examinations were also carried out at the same designated times in the children of the control group.

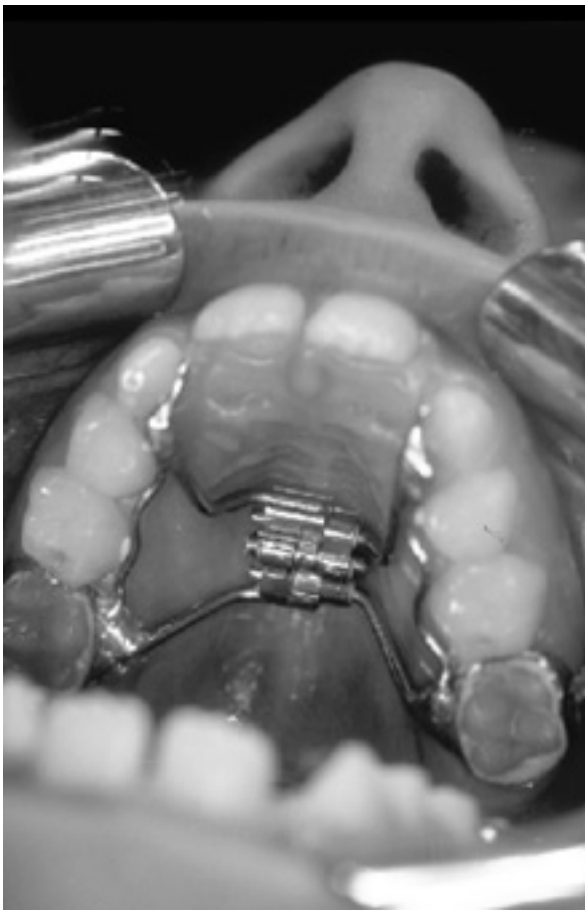

Figure 1. Rapid maxillary expansion appliance.

None of these children have been submitted to any other otolaryngologic or dental surgery during the study period.

During the study four children, three from the control group and one from the experimental group, were excluded (Figure 2).

Statistics

Descriptive statistics were obtained for acoustic rhinometry data at T0, T1 and T2, for both the RME and control groups. Within each of the treatment groups, for the different variables under study, the effect of time was analysed using the nonparametric Friedman test and individual pairwise comparisons were done with the nonparametric Wilcoxon test. All statistical analysis was performed using SPSS version 11.0 for Windows.

RESULTS

According to general recommendations we only considered the measurements taken after vasoconstriction [12]. The individual results are shown in Table 1.

Although the experimental design has considered different age groups, the sample size is too small to allow reliable analysis of this factor effect.

Total nasal volume (TV), minimal cross-sectional area (MCA),

Table 1. Distribution of studied population; F – female, M – mail, TV – total nasal volume, MCA – minimal cross-sectional area, D – distance at witch this occurred, n.a. – not acquired.

Code number	Gender	Age	Group	TV at T0 [cm^3]	TV at T1 [cm^3]	TV at T2 [cm^3]	MCA at T0 [cm^2]	MCA at T1 [cm^2]	MCA at T2 [cm^2]	D at T0 [cm]	D at T1 [cm]	D at T2 [cm]
1	F	6	Expanded	5.74	12.15	8.67	0.45	0.52	0.49	0.66	0.42	0.54
2	M	5	Expanded	5.01	9.92	9.66	0.39	0.39	0.48	0.42	0.54	0.54
3	M	4	Control	5.35	5.98	5.31	0.47	0.41	0.46	1.98	0.30	0.78
4	F	6	Control	7.20	7.63	6.41	0.50	0.40	0.48	0.54	0.66	0.78
5	F	8	Expanded	5.62	8.73	8.53	0.40	0.33	0.42	0.30	0.66	0.66
6	F	9	Expanded	8.88	7.49	5.88	0.68	0.44	0.42	0.30	0.54	2.10
7	M	7	Expanded	4.99	5.59	7.54	0.37	0.42	0.50	2.10	0.66	0.42
8	M	8	Expanded	4.27	5.56	6.04	0.31	0.42	0.39	0.90	0.42	0.42
9	F	7	Expanded	6.30	10.67	9.06	0.42	0.59	0.54	0.78	0.42	0.42
10	F	8	Control	7.51	7.71	12.07	0.64	0.49	0.62	0.30	0.42	0.30
11	F	7	Control	8.14	15.10	11.39	0.51	0.66	0.52	0.42	0.12	0.42
12	M	9	Control	10.03	6.12	15.63	0.49	0.43	0.62	0.78	1.02	0.30
13	M	8	Control	7.59	8.62	9.80	0.42	0.44	0.45	0.54	0.54	0.54
14	M	11	Expanded	7.01	5.45	n.a.	0.44	0.27	n.a.	0.42	0.30	n.a.
15	M	10	Expanded	6.80	7.96	10.45	0.48	0.56	0.53	1.26	0.30	0.42
16	F	10	Expanded	6.75	7.19	7.33	0.51	0.53	0.53	1.14	1.14	0.54
17	F	12	Expanded	5.13	9.41	9.28	0.45	0.64	0.67	2.10	0.18	0.12
18	M	11	Expanded	5.76	7.11	7.17	0.39	0.45	0.49	1.02	0.42	0.42
19	F	10	Expanded	5.17	6.73	5.70	0.34	0.36	0.41	0.66	0.54	0.42
20	F	12	Control	7.26	5.00	6.44	0.38	0.20	0.37	0.42	1.02	0.90
21	F	12	Control	10.82	9.25	8.94	0.47	0.40	0.51	0.18	0.42	0.30
22	F	12	Control	7.82	9.23	14.26	0.45	0.47	0.61	0.42	0.78	0.30
23	F	12	Control	8.21	3.55	7.04	0.57	0.21	0.54	0.78	1.14	0.42
24	M	11	Control	6.49	6.10	n.a.	0.64	0.61	n.a.	0.18	0.78	n.a.

Table 2. Acoustic parameters: mean and standard deviation at each of the experimental stages (sample size).

	group	T0	T1	T2
TV	RME	$5.96^{a} \pm 1.20$ (13)	$8.00^{b} \pm 2.08$ (13)	$7.94^{b} \pm 1.57$ (12)
[cm^3]	Control	7.86 ± 1.51 (11)	7.66 ± 3.05 (11)	9.73 ± 3.54 (10)
MCA	RME	$0.43^{a} \pm 0.09$ (13)	$0.45^{a,b} \pm 0.11$ (13)	$0.49^{b} \pm 0.08$ (12)
[cm^2]	Control	0.50 ± 0.08 (11)	0.43 ± 0.14 (11)	0.52 ± 0.08 (10)
D	RME	0.93 ± 0.59 (13)	0.50 ± 0.24 (13)	0.58 ± 0.49 (12)
[cm]	Control	0.60 ± 0.50 (11)	0.65 ± 0.33 (11)	0.50 ± 0.23 (10)

T0 – time immediately before the application of the intra-oral device, T1 - time immediately after the end of rapid maxillary expansion (approximately one month after T0), T2 - time after almost 5 months of the retention period; TV - total nasal volume, MCA - minimal cross-sectional area, D - the distance at which this occurred; $^{a, b}$ – homogeneous groups according to the Wilcoxon test ($p<0.05$).

and the distance (D) at which this occurred was computed as the average obtained from both nasal sides. The results are shown in Table 2.

On average, children undergoing expansion showed a significant increase in the total nasal volume from T0 to T1 that persisted through T2. No significant difference was observed after removing the appliance. Children in the control group did not show any significant changes during the study period.

When considering the relative gain of the total nasal volume, there was a significant difference ($p<0.05$) between the RME group and the control group from T0 to T1, but not from T0 to T2 (Figure 3).

Comparing the total nasal volume change between T0 and T2, 92 % (11) of the RME group showed an increase and only 8% (1) revealed a negative result. In the control group half of the children demonstrated an increased volume whereas the other half had a decreased volume. Results from the Fisher Exact Test showed that the intraoral expansion device resulted in a significant rise in the proportion of children with increased total nasal volume ($p<0.05$). Regarding the evolution of the

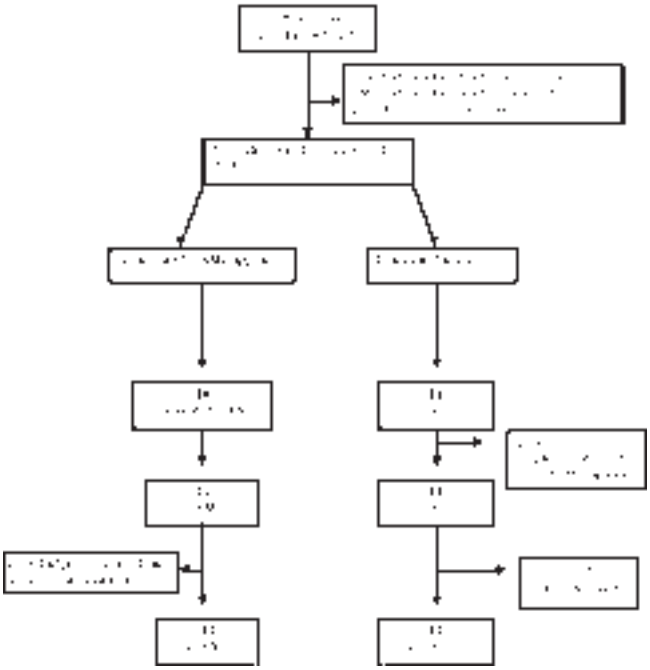

Figure 2. Study profile. DS - Down syndrome, RME - Rapid maxillary expansion group, controls – group without treatment, T0 – time immediately before the application of the intra-oral device, T1 - time immediately after the rapid total expansion (approximately one month after T0), T2 - time after the retention period, almost 6 months after T0.

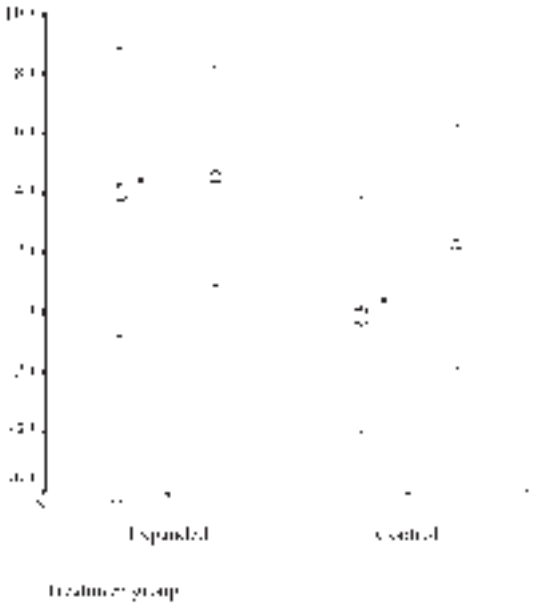

Figure 3. Means and standard deviations of proportional nasal volume change in relation to the initial volume at T0 (%) for both groups: "○" - from T0 to T1 and "□" - from T0 to T2. Differences according to the nonparametric Mann-Whitney test (* - $p<0.05$).

MCA over time, a significant difference was noticed for children undergoing expansion (Figure 4).

The distance from the minimal cross-sectional area did not show any significant difference at various experimental stages in either experimental or control groups. Nevertheless, in the RME group

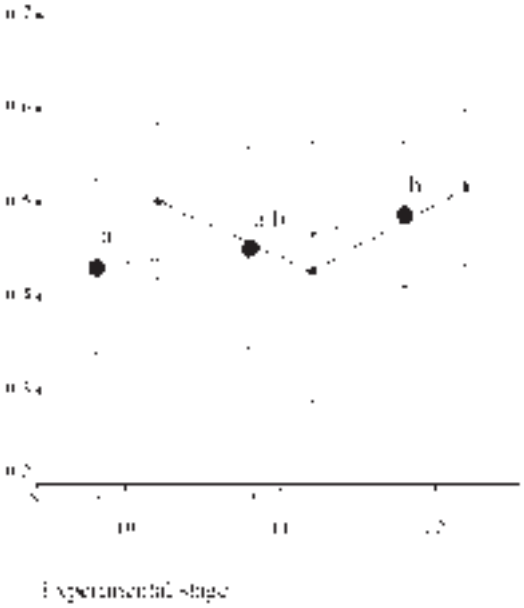

Figure 4. Means and standard deviations of MCA (cm^2) after vasoconstriction at each of the experimental stages (N - sample size): "•"-expanded group; "*" - control group. [a,b] – homogeneous groups according to the Wilcoxon test ($p<0.05$).

a noticeable change was observed between T0 and T1, with almost no change at the last stage (T2). In contrast, results from the control group did not show any significant change over time.

DISCUSSION

The present study evaluated the effects of RME on nasal patency in a group of children with DS. This form of treatment is well established in the general paediatric population[5, 6, 7, 8].

It is of note that the sample size is small as a result of the selection criteria and the difficulty to pursue a prospective clinical investigation over an extended time period.

Some authors consider RME as a medical treatment for nasal obstruction, recurrent ear and nose infections in the general paediatric population[13]. Our selected group of children with DS presented both clinical symptoms such as nasal obstruction and/or recurrent ear and nose infection as well as signs of dental cross bite.

Beyond the positive dental improvement, there are also skeletal effects. The greatest movement from RME is inferiorly and anteriorly. The separated palatine bones widen the maxilla and there is often some splaying of the pterygoid process of the sphenoid bone. The lateral walls of the nasal airflow incline outwards taking with them the inferior turbinate bones and enlarging the airway[14, 15]. The retention period, with nearly 5 months of duration, consisted in the maintenance of the intra-oral appliance after the end of active expansion. This period leads to a normal radiographic appearance of the midpalatal suture evident three months after expansion and three months later, the reestablishment of a normal histologic appearance of the suture[16, 17]. As the nasal cavity is high and narrow, a small increase in width produces an augmentation in the cross

sectional area and permits the passage of a vastly increased volume of air. The gain in the total nasal volume and in the MCA obtained in children of the RME group is most likely due to these skeletal changes. Comparing the RME and the control groups, the results are more evident when considering the relative gain in the total nasal volume, with a significant difference on the evolution observed after maximal expansion (T0 to T1). Moreover, the results show that over the treatment period (T0 to T2) a significantly larger proportion of the children undergoing expansion presented an increased nasal volume when compared with those from the control group. Generally, over short periods of time, fluctuation in total nasal volume can be expected due to changes of densities, viscosities and congestion of the nasal mucosa or to the exudate present[18, 19]. This effect may not be completely reversed by application of a vasoconstrictor. Moreover, the observed decreased volume between different observational times is in general very reduced for controls and negligible for the expanded group. Probably, the volume gain promoted by the maxillary expansion is so significant that it dominates over this effect.

Thus, these results may express the possible initial overexpansion of the maxillary bone produced by standardized RME. The stability of the increase in nasal width after RME has been reported in several studies[4,20]. Long-term effects of RME appear to involve a portion of the craniofacial complex with enhanced transverse growth of the circum-maxillary anatomical regions[4].

Several investigations described the reduction of nasal airway resistance after RME using active rhinometry [4, 5, 7, 8]. In our study we assessed the nasal airway by acoustic rhinometry which is known to be an objective method of assessing this area as a function of distance and hence provides a geographic description of the nasal cavity. It is a simple, rapid and noninvasive technique and requires minimal cooperation [10]. These attractive features are relevant to its application in paediatric populations, especially those with mental retardation.

This is the first known study using rapid maxillary expansion in children with DS and these are the first data of acoustic rhinometry of this population.

According to the literature concerning RME [4, 5, 6, 7, 8, 13, 14, 15] and the phenotypic characteristics of Down syndrome, these children should be considered for RME in order to correct some of the observed cranial skeletal deformities of the syndrome. In children with DS this procedure permits the widening of the maxillary bones with a gain in nasal volume and also gives more space for the tongue in the oral cavity.

In conclusion, RME provides a significant gain of nasal volume and minimal cross-sectional area in children with DS. This procedure may be used in combination with other treatments for nasal obstruction and apnoea syndromes.

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REFERENCES

1. Fryns JP (1990) In: Birth Defect Encyclopedia. Mary Louise Buyse, Editor in Chief, Blackwell Scientific Publication, Inc., pp.391-393.
2. Mastroiacovo P, Bertollini R, Corchia C (1992) Survival of children with Down syndrome in Italy. American Journal of Medical Genetics 42: 208-212.
3. Gorlin R, Cohen MM, Levin LS (1990) Chromosomal Syndromes: Common and/or Well-Know Syndromes: Trisomy 21 syndrome (Down Syndrome) In: Syndromes of the head and neck. 3rd edit. New York: Oxford University Press, pp 33-40.
4. Cameron CG, Franchi L, Baccetti T, MacNamara (2002) Long term effect of rapid maxillary expansion: a posteroanterior cephalometric evaluation. J. Am J Orthod Dentofacial Orthoped 121: 129-135.
5. Hershey HG, Stewart BI and Warren DW (1976) In: Changes in nasal airway resistance associated with rapid maxillary expansion. Am J Orthod 69: 274-284.
6. Hartgerink DV, Vig PS and Abbott DW (1987) The effect of rapid maxillary expansion on nasal airway resistance. Am J Orthod Dentofacial Orthop 92: 381-389.
7. White BC, Woodside DG and Cole P (1989) The effect of rapid maxillary expansion on nasal airway resistance. J Otolaryngol 18: 137-143.
8. Wollens AG, Goffart Y, Lismonde P and Limme M (1991) Therapeutic maxillary expansion. Rev Belge Med Dent 46: 51-58.
9. Hilberg O, Jackson AC, Swift DL, Pedersen OF (1989) Acoustic rhinometry: evaluation of nasal cavity geometry by acoustic reflection. J Appl Physiol 66: 295-303.
10. Djupesland P, Pedersen OF (2000) Acoustic rhinometry in infants and children. Rhinol Suppl 16: 52-58.
11. Andrade DC (2000) Protrusão Lingual e Placas Palatins na Infância. Faculdade de Medicina Dentária da Universidade do Porto. PhD Thesis, pp 98-101.
12. Grymer L (2000) Clinical applications of acoustic rhinometry. Rhinol Suppl 16: 35-43.
13. Gray LP (1975) Results of 310 cases of rapid maxillary expansion selected for medical reasons. J Laryngol Otol 89: 601-614.
14. Timms DJ (1980) A study in basal movement with rapid maxillary expansion: Am J Orthod 77: 500-5007.
15. Timms DJ (1984) The reduction of nasal airway resistance by rapid maxillary expansion and its effects on respiratory disease. J Laryngol Otol 98: 357-362.
16. Stambuch HK, Cleall JF (1964) The effects of splitting the midpalatal suture on the surrounding structures. Am J Orthod 50: 923-924.
17. Cleall JF, Bayne DI, Posen JM (1965) Expansion of midpalatal suture in the monkey. Angle Orthod 35: 23-35.
18. Hilberg O, Pedersen OF, Jensen FT (1994) Nasal patency evaluated by acoustic rhinometry and magnetic resonance imaging. Am J Rhinol 8: 290-291.
19. Corey JP, Kemker BJ, Nelson R, Gungor A (1997) Evaluation of the nasal cavity by acoustic rhinometry in normal and allergic subjects. Arch Otolaryngol Head Neck Surg 117: 22-28.
20. Werts R, DreskinM (1977) Midpalatal suture opening: a normative study. Am J Orthod 71: 367-381.

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