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
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 otolaryngologi-
cal 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 com-
pression, (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 ran-
domly assigned to two groups: RME and control. They under-
went acoustic rhinometry which was always performed by the
same examiner. The instrument used for these examinations
was an Eccovision Acoustic Rhinometer with a software ver-
sion 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 car-
ried out at the same designated times in the children of the
control group.

None of these children have been submitted to any other oto-
laryngologic 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 non-
parametric 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 indi-
vidual 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),
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

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

<table>
<thead>
<tr>
<th>Code number</th>
<th>Gender</th>
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<th>Group</th>
<th>TV at T0 [cm³]</th>
<th>TV at T1 [cm³]</th>
<th>TV at T2 [cm³]</th>
<th>MCA at T0 [cm²]</th>
<th>MCA at T1 [cm²]</th>
<th>MCA at T2 [cm²]</th>
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<td>7.11</td>
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<td>0.61</td>
<td>n.a.</td>
<td>0.18</td>
<td>0.78</td>
<td>n.a.</td>
</tr>
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</table>

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

<table>
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<th>group</th>
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<th>T1</th>
<th>T2</th>
</tr>
</thead>
<tbody>
<tr>
<td>TV</td>
<td>5.96 ± 1.20 (13)</td>
<td>8.00 ± 2.08 (13)</td>
<td>7.94 ± 1.57 (12)</td>
</tr>
<tr>
<td>[cm³]</td>
<td>7.86 ± 1.51 (11)</td>
<td>7.66 ± 3.05 (11)</td>
<td>9.73 ± 3.54 (10)</td>
</tr>
<tr>
<td>MCA</td>
<td>0.45 ± 0.09 (13)</td>
<td>0.45 ± 0.11 (13)</td>
<td>0.49 ± 0.08 (12)</td>
</tr>
<tr>
<td>[cm²]</td>
<td>0.50 ± 0.08 (11)</td>
<td>0.43 ± 0.14 (11)</td>
<td>0.52 ± 0.08 (10)</td>
</tr>
<tr>
<td>D</td>
<td>0.93 ± 0.59 (13)</td>
<td>0.50 ± 0.24 (13)</td>
<td>0.58 ± 0.49 (12)</td>
</tr>
<tr>
<td>[cm]</td>
<td>0.60 ± 0.50 (11)</td>
<td>0.65 ± 0.33 (11)</td>
<td>0.50 ± 0.23 (10)</td>
</tr>
</tbody>
</table>

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; 

- homogeneous groups according to the Wilcoxon 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 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 intraoral 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

Figure 4. Means and standard deviations of MCA (cm²) 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.
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