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Development of craniofacial and dental arch morphology in relation to sleep disordered breathing from 4 to 12 years. Effects of adeno-tonsillar surgery.

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Running title:

Dental arch morphology and sleep disordered breathing

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Abstract

Objectives: To study the development of craniofacial and dental-arch morphology in children with sleep disordered breathing in relation to adeno-tonsillar surgery.

Subjects and method: From a community based cohort of 644 children, 393 answered questionnaires at age 4, 6 and 12 years. Out of this group, twenty-five children who were snoring regularly at age 4 could be followed up to age 12 together with 24 controls not snoring at age 4, 6 and 12 years. Study casts were obtained from cases and controls and lateral cephalograms from the cases. Analysis regarding facial features and dento-alveolar development was performed.

Results: Children snoring regularly at age 4 showed reduced transversal width of the maxilla and more frequently had anterior open bite and lateral cross-bite than the controls. These conditions persisted for most cases at age six, by which time 18/25 had been operated for snoring. In most of the cases, surgery cured the snoring temporarily, but their width of the maxilla was still smaller by age 12—even when nasal breathing was attained. At age 12, the frequency of lateral cross-bite was much reduced and anterior open bite was resolved, both in cases and controls. The children who snored regularly at age 12 showed a long face anatomy and were oral breathers; this applied even to those who were operated. The seven cases who were not operated and the five who were still snoring in spite of surgery at age 12, did not have reduced maxillary width as compared to the controls.

Conclusion: Dento-facial development in snoring children is not changed by adenotonsillar surgery regardless of symptom relief.

Key Words:

Snoring, sleep disordered breathing, dental arch morphology, surgery, adenoidectomy, tonsillectomy, development.

Introduction

More than a century ago, ever since the Danish otorhinolaryngologist Wilhelm Meyer in 1868 [1] described the effects of adenoidectomy, it has been pointed out that children with sleep related breathing difficulties often show a facial appearance different from that of children without these problems. However, the debate is still ongoing as to whether this "long face" condition is partly caused by the breathing obstruction, the hypertrophy of the adenoids and/or tonsils, or if this development is solely genetically based.

In 1969, Moss wrote about "the primary role of functional matrix in facial growth." explaining the mechanism behind the influence of environmental forces [2].

Solow demonstrated that individuals snoring regularly and OSA patients have a deviant posture of their head and neck compared to non-snoring controls [3]. He also pointed out the pressure on the growing facial skeleton by "soft tissue stretching" [4]. Experimentally induced nasal obstruction in rhesus monkeys has been shown to lead to immediate alteration of head/neck posture to facilitate oral breathing (Miller 1984) [5]. Permanent induced obstruction has led to altered facial development in growing primates as shown by Harvold 1968 [6]. On the other hand, Harterink & Vig (1989) [7] among others, did not find any connection between breathing obstruction and facial anatomy.

Over recent decades, obstructive adenoids and tonsils have been shown to be associated with specific anatomical traits that seem to be at least partly reversible by surgery [8–10]. All of these previous studies have dealt with groups of selected patients who have sought treatment for their breathing obstruction. Morphological studies of obstructed breathing in children have not been explored in connection to community-based cohort studies with long-term follow-up.

We have earlier described the epidemiology of snoring and the dento-facial features connected to their symptoms in a cohort of four-year-old children who were followed up to age 12 [11–13].

The purpose of the present study was to investigate the long term effect of early surgery for sleep disordered breathing. This was done by following each child in the cohort who was snoring regularly at age four with respect to obstructive symptoms and dento-alveolar and craniofacial anatomy, before and after surgery up to age 12.

Subjects and methods

This study was approved by the ethical committee of Uppsala University and informed consent was obtained from all the parents.

Subjects

As part of the Swedish national community health program, children have a dental examination at age four. Over a period of 16 consecutive months, all four-year-old children in a small town received a questionnaire concerning snoring and related problems in connection to the dental examination. Of a total of 644 children, 615 (95.5%) answered the questionnaire at age four. When those 615 children reached age six, they were invited to the next examination. Five hundred and nine children took part in this first follow-up and were again contacted six years later to participate in a second follow-up at age twelve. Three hundred and ninety three children participated in all three examinations [11–13].

The present investigation is a case-control study from the 393 children answering the questionnaire at all of the ages four, six and twelve. The *cases* consist of all children who were snoring regularly at age four, who could be followed individually, including with study models, up to age twelve, (n=25, 8 boys and 17 girls). Serving as *controls* were the 24 children (11 boys and 13 girls), from the original control group of 48 children not snoring at age 4 [11] who did not later report snoring either at age 6 or 12 and also could be followed with study models.

None of the children included in the study had completed or ongoing orthodontic treatment.

Methods

Questionnaires

The questionnaires given at four, six and twelve years included questions concerning the child's snoring and apneas, sleeping habits, oral breathing, sucking habits and other related conditions. See table 1. For details see [13].

Table 1: Symptoms at age 4, 6 and 12 in CASES, in comparison to symptoms in CONTROLS at corresponding ages.

	Cases, *n* = 25			**Controls, *n* = 24**		
Age in years	4 years	6 years	12 years	4 years	6 years	12 years
SEN	25	7	10	0	0	0
SS	0	10	8	0	0	0
NS	0	8	7	24	24	24
Apneas	11	3	1	0	0	0
Restless sleep	0	0	2	0	0	0
Oral breathing every night	12	2	7	0	0	0
Prolonged sucking habits	17	7	1	14	7	1
Asthma			4			0
Allergy			3			0
Other health problems			3			0
Other family member snoring			19			6

ENT and sleep examinations

At age four, all *cases* underwent an ENT examination followed by a sleep polygraphy [11]. In the two follow up studies at six and twelve years, they underwent new clinical ENT examinations. Those cases that were also snoring every night at age 12 were given a second sleep polygraphy as were six of the not-snoring controls [13].

Orthodontic examinations

Biometric assessments.

All *cases* and *controls* had study models taken at age 4, 6 and 12. The biometric variables recorded were: Angle class (sagital relations between the dental arches), anterior open bite, lateral cross-bite, overjet, overbite, length of the upper and lower arches, width of the maxilla and mandible as measured between the cusp tips of the deciduous or permanent canines, and between the mesio-lingual cusps in the first and second deciduous molars or the first and second permanent premolars, according to Moorrees et al (1969) [14] (fig. 1 a and b). Orthodontic diagnoses were analyzed (see table 2). Measurements of maxillary width were studied for boys and girls separately [15].

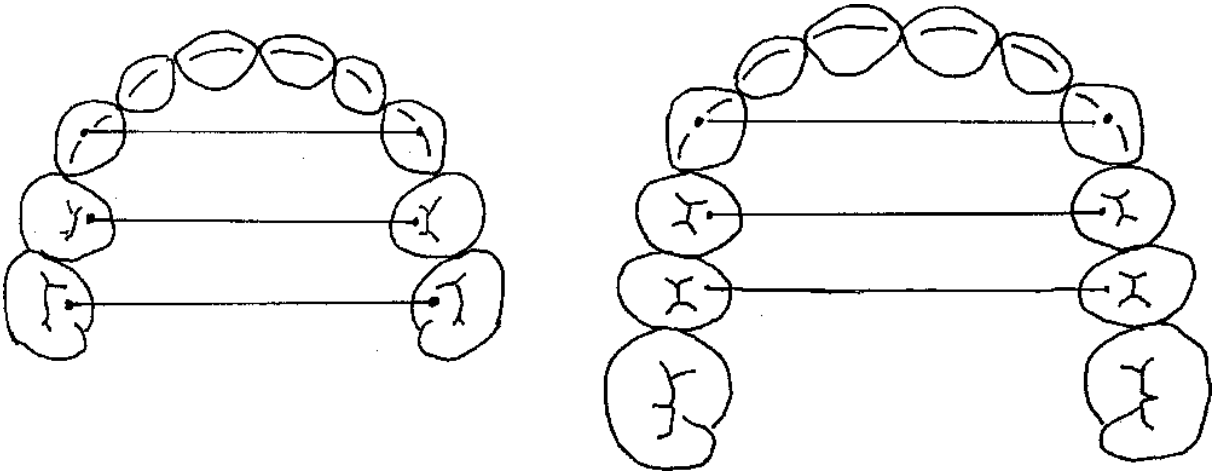

Figure 1: Dental arch measurements for primary and permanent dentition of the maxilla.

Dental arch measurements were done on persisting deciduous teeth or fully erupted permanent teeth. A sliding caliper with 0.01 mm accuracy was used for measuring distances.

Table 2: Definitions of orthodontic diagnoses.

Angle class 1	The first deciduous molars or first permanent molars in neutral relation on both sides.
Angle class 2	The first deciduous molars or first permanent molars in post-normal relation in one or both sides.
Angle class 3	The first deciduous molars or first permanent molars in pre-normal relation in one or both sides.
Lateral cross-bite	Two or more teeth in one or both sides in inverted occlusion (the buccal cusps of the upper teeth occluding lingually of the buccal cusps of the lower teeth).
Anterior open bite	Vertically open bite between all incisors when the molars are occluding.

Radiographic assessments

Lateral cephalograms were obtained from 18 of the cases at age four. 13 of those children had follow-up radiographs examinations done at age 6. The seven cases still snoring regularly at age 12 were once again examined cephalmetrically.

The cephalograms were taken in a standardized manner with the child's head fixed in a cephalostat with the film parallel to the profile and the head orientated in a natural position. The teeth were in intercuspal position. The cephalometric reference points and lines used are those defined by Björk (1947) [16] (fig 2). The cephalometric landmarks were digitized and analyzed by the first author. Different cephalostats were used, either in an orthodontic office or in the hospital, but no adjustment for enlargement was done as only angles and ratios were calculated.

The controls were not assessed with lateral cephalograms as current Swedish regulations do not allow radiographic examination of non-symptomatic subjects. Instead, normative data from Swedish material [17] was used for comparison.

Surgery

Adenoidectomy (A) and/or tonsillectomy (T) was performed in 18 of the 25 cases, all but one between the ages of four and six years. The choice of surgical procedure depended on the

findings during ENT-examination; small tonsils were not removed. Seven children were not operated due to parental reluctance

Statistics

Student's t-test was used for parametric data and for comparison of data between cases and controls and Fisher's Exact Test when small numbers were included. χ^2 was used for categorical data. Descriptive data are used to present the effect of surgery for individual children. Rational data was expressed as means, standard deviation, and range.

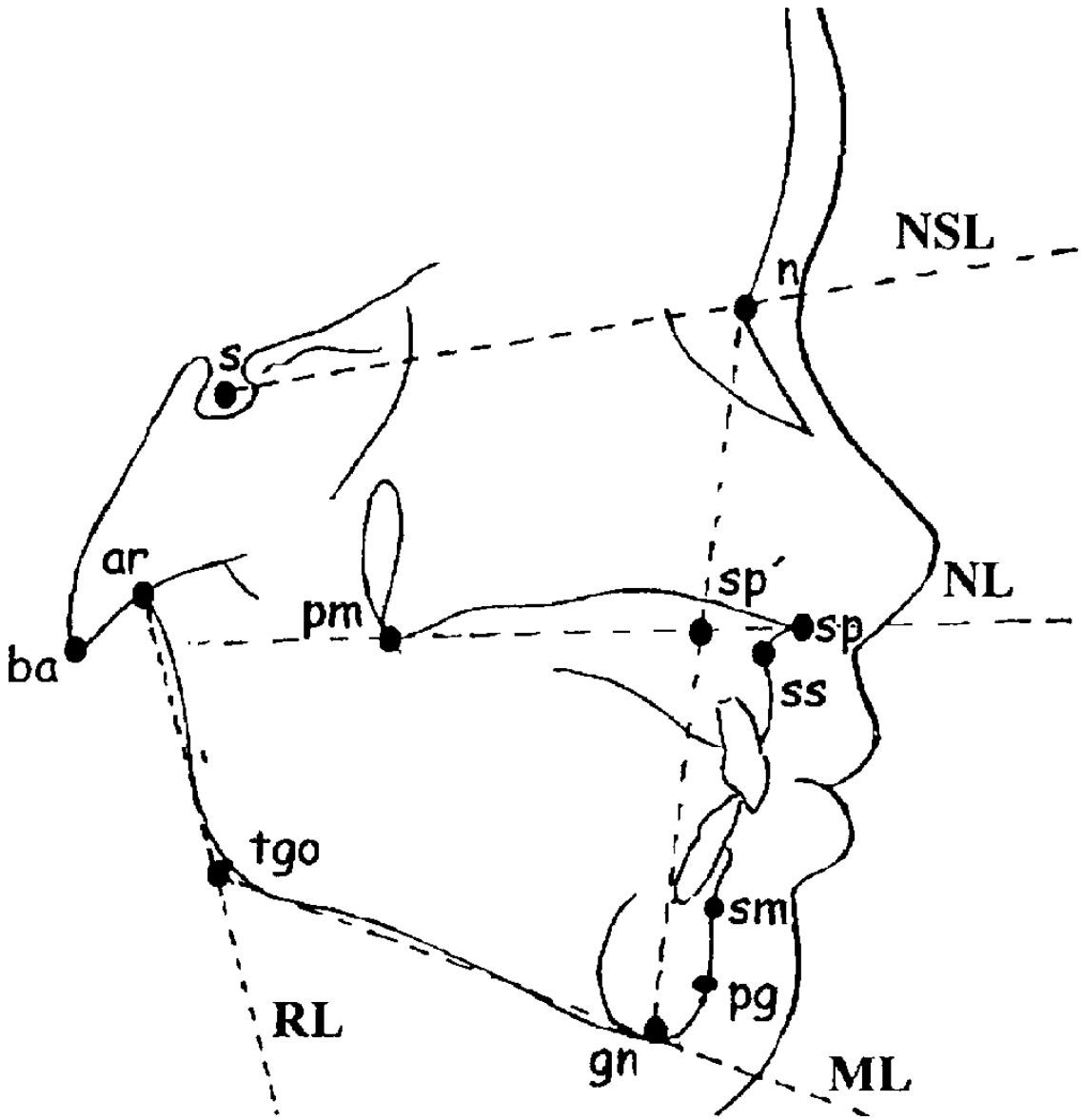

Figure 2: Reference points and lines used in the cephalometric analyzes, according to Björk [17].

Error of methods

Duplicate independent determinations for biometric and cephalometric measurements for a sub-sample of 15 models and radiographs were calculated according to the formula

$$S_e = \sqrt{\frac{\Sigma(a_2 - a_1)}{2n}}$$

where a_1 =the first measurement, a_2 = the second, and n= the number of duplicates.

Results

Answers to the questionnaires in the case and control groups

The symptoms (answers to the questionnaires) at age 4, 6 and 12 for the cases and the not-snoring controls are presented in Table 1. Between age 4 and 6, by which time 18/25 of the cases had been operated, a decrease in the number of children reporting snoring is seen and less than half of those still snoring at age 6 were snoring regularly. The number of children reporting oral breathing at age 6 had decreased as well. At age 12, 18/25 of the cases were snoring and the number with oral breathing had increased again. Prolonged sucking habits (≥3 years, ≥5 years and 12 years) were twice as common at age 4 compared to age 6, both for cases and controls, but the number did not differ significantly between the groups at any age. A significant difference between the groups was that the cases reported more snoring among family members than did the controls ($p < 0.00054$).

The individual development of snoring among the cases is described in relation to type of surgery in Fig 3, where oral breathing at age 12 also is shown. At the age of six—after surgery—all but 4/18 operated children had either stopped snoring (8/18) or snored “sometimes”(6/18). All not operated cases were still snoring at age six, 3/7 every night and 4/7 “sometimes”. At age 12, five of the operated children who were cured from snoring at six years had started to snore again; one of them (operated with A+T) was snoring every night. Seven of the 18 operated cases (5 given A+T or T and 2 solely A) did not snore at age 12—nor did 3/7 of those not operated. At age 12, all children snoring every night were breathing orally, most of them “always”, and half of the ones not snoring were

never breathing orally. Snoring at age 12 was not significantly different between operated (n=18) and not operated cases (n=7).

Biometrics

The orthodontic diagnoses at age 4, 6 and 12 for cases and controls are shown in table 3, where the operated cases are divided into “cured” (not snoring) and “not cured”. Anterior open bite and lateral cross-bite were more common among the cases than among the controls at age 4 before surgery and also at age 6 after surgery. At age 12, anterior open bite had been resolved for all children except for one of the controls. At age 12, a few examples of lateral cross-bite were still seen, more commonly among the operated “cured” children (4/13) than among the controls (2/24).

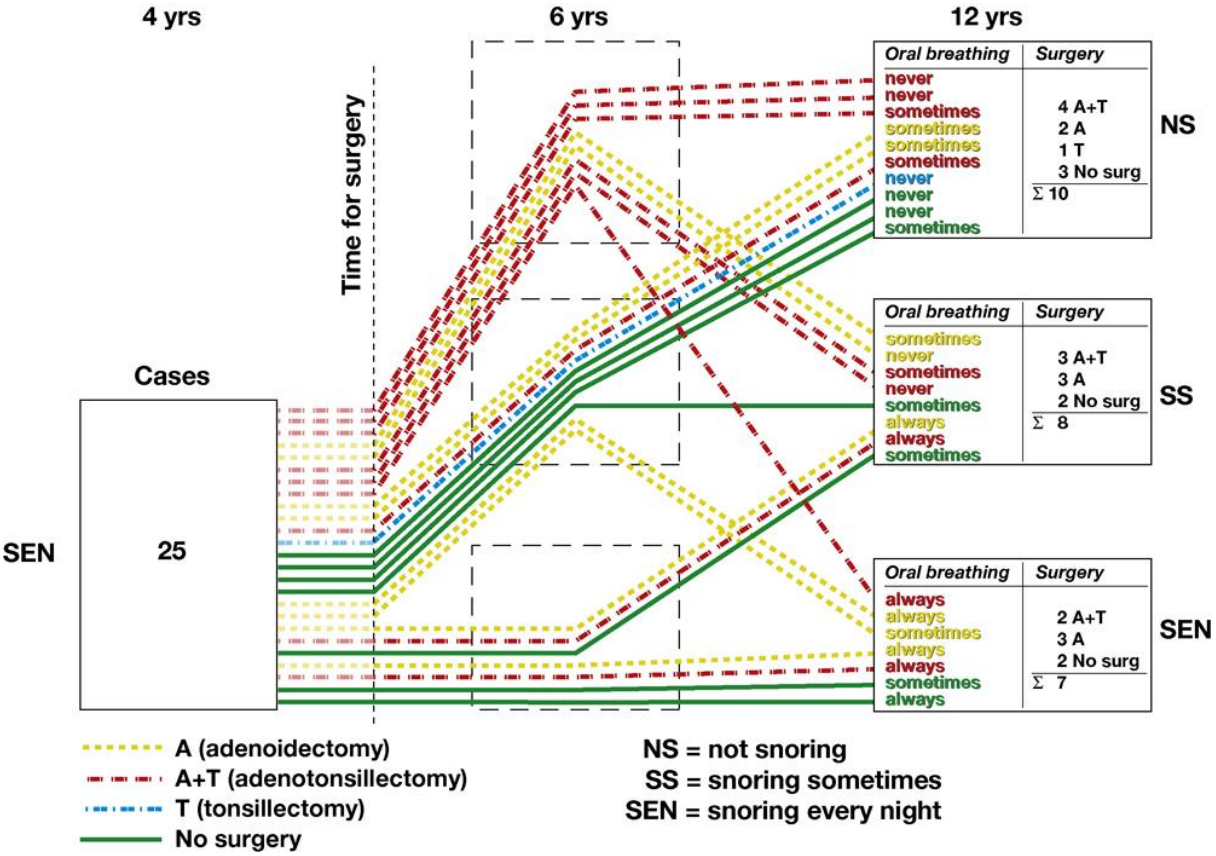

Figure 3: Individual development of snoring among the cases from 4 to 6 to 12 years, in relation to type of surgery or no surgery, and notion on oral breathing at age 12.

Table 3: The development of orthodontic diagnoses among operated and not operated cases and controls, from 4 to 6 to 12 years.

Age	**Operated "cured" at 12 years, *n* = 13**	**Operated "not cured" at 12 years, *n* = 5**	**Not op., *n* = 7**	**Controls, *n* = 24**	**Level of sign**
Post-normal relation					
4 years	2	0	0	3	n.s.
6 years	3	0	0	5	n.s.
12 years	6	1	0	2	n.s.
Lateral cross-bite					
4 years	9	1	1	3	*
6 years	10	2	1	3	**
12 years	4	0	1	2	n.s.
Anterior open bite					
4 years	12	3	0	7	***
6 years	7	3	0	6	*
12 years	0	0	0	1	*

* $p < 0.05$.

** $p < 0.01$.

*** $p < 0.001$.

Upper and lower arch-length, overbite and over jet, width in the lower jaws at canines and first and second (pre-) molars were measured and analyzed for both cases and controls. No statistical differences were found between the groups for any measurements except for maxillary width.

The measurements of maxillar width at canines and first and second molar/premolar at ages 4, 6 and 12 in the 25 cases and the 24 controls for boys and girls, are shown in Table 4.

There is a difference between cases and controls at all ages. This difference reaches the level of significance for the girls, but not for boys (small numbers). Before surgery, at age four, the cases had a more narrow upper jaw than the controls at the primary canines and first and

second primary molars. This difference remained both at age 6 and 12 when 18/25 cases were operated.

Table 4: Measurements of maxillary width at ages 4, 6 and 12 for cases, of whom 18/25 were operated between ages 4 and 6, and controls.

Distances in mm	**Age**	**Cases, *n* = 25**		**Controls *n* = 24**		**Level of significance**	
		♂ = 8	**♀ = 17**	**♂ = 13**	**♀ = 11**	♂	♀
Inter-canin width	4 years	28.4 ± 1.51	25.5 ± 2.58	29.2 ± 2.88	28.7 ± 2.89	n.s.	***
	6 years	29.7 ± 1.48	27.3 ± 2.63	29.9 ± 2.93	30.1 ± 2.84	n.s.	**
	12 years	33.7 ± 1.71	31.1 ± 2.15	34.1 ± 2.58	32.9 ± 3.04	n.s.	*
Inter-first (pre-)molar width	4 years	28.4 ± 2.37	25.8 ± 2.24	28.6 ± 2.92	28.1 ± 2.79	n.s.	**
	6 years	29.2 ± 2.19	26.7 ± 2.22	29.3 ± 2.88	28.7 ± 2.89	n.s.	*
	12 years	30.3 ± 2.09	27.1 ± 2.68	31.7 ± 3.52	29.5 ± 4.08	n.s.	n.s.
Inter-second (pre-)molar width	4 years	33.5 ± 2.72	30.7 ± 2.23	33.2 ± 3.03	32.3 ± 2.06	n.s.	*
	6 years	34.3 ± 2.93	31.8 ± 2.14	34.4 ± 2.63	33.2 ± 2.32	n.s.	*
	12 years	35.3 ± 2.57	32.5 ± 2.67	36.5 ± 2.71	35.0 ± 3.20	n.s.	n.s.

* $p < 0.05$.

** $p < 0.01$.

*** $p < 0.001$.

The result of surgery was further analyzed by splitting the cases into sub-groups: “operated cured” (not snoring), “operated not cured”, and “not operated” (Table 5).

At all ages, the maxillary width was smaller among the operated “cured” cases than among the controls at all points of measurement. This difference reaches the level of significance for the girls, but not for boys (small numbers). The cases “not cured” at 12 and the cases not operated upon had almost the same measurements as the controls.

As compared to the controls, it can be seen that the operated cases had greater transverse development from age four to six between the canines, but not between the molars (Table 5).

Table 5: Measurements of maxillary width at ages 4, 6 and 12 for cases, in relation to effect of surgery between 4 and 6, or no surgery, and controls. Significant differences noted between "operated cured" girls and girls in the control group.

		Case operated "cured" at 12 years, *n* = 13		**Case operated "not cured" at 12 years, *n* = 5**		**Case not operated, *n* = 7**		**Controls, *n* = 24**		**Level of sign. cases op cured vs controls**
Maxillary width in mm	**Age in years**	**♂ *n* = 3**	**♀ *n* = 10**	**♂ *n* = 1**	**♀ *n* = 4**	**♂ *n* = 4**	**♀ *n* = 3**	**♂ *n* = 13**	**♀ *n* = 11**	**♀**
Inter-canin	4	27.5 ± 1.50	24.9 ± 2.18	29.9	26.4 ± 3.95	28.8 ± 1.49	26.1 ± 4.16	29.2 ± 2.88	28.7 ± 2.89	***
	6	29.1 ± 1.22	26.9 ± 2.07	31.3	28.2 ± 2.87	29.7 ± 1.71	27.3 ± 4.42	29.9 ± 2.93	30.1 ± 2.84	***
	12	33.8 ± 0.24	30.3 ± 1.96	35.7	33.0 ± 1.93	33.2 ± 2.26	32.8 ± 1.53	34.1 ± 2.58	32.9 ± 3.04	**
Inter-first (pre)mol.	4	26.8 ± 1.96	25.3 ± 1.81	31.1	26.8 ± 2.25	29.0 ± 2.29	26.1 ± 3.76	28.6 ± 2.92	28.1 ± 2.79	**
	6	27.3 ± 1.60	26.1 ± 1.82	31.8	28.4 ± 2.25	30.0 ± 1.75	26.8 ± 3.12	29.3 ± 2.88	28.7 ± 2.89	**
	12	29.4 ± 1.69	25.8 ± 2.18	32.7	28.4 ± 2.67	30.4 ± 2.36	29.6 ± 2.09	31.7 ± 3.52	29.5 ± 4.08	**
Inter-sec. (pre-)mol.	4	31.4 ± 1.05	30.2 ± 1.92	36.9	31.7 ± 1.84	34.3 ± 2.81	30.9 ± 3.91	33.2 ± 3.03	32.3 ± 2.06	*
	6	32.0 ± 2.04	31.4 ± 1.64	37.3	32.8 ± 1.95	35.4 ± 2.69	32.0 ± 3.98	34.4 ± 2.63	33.2 ± 2.32	**
	12	34.5 ± 2.60	31.7 ± 2.45	37.2	34.0 ± 3.24	35.5 ± 2.99	33.1 ± 2.29	36.5 ± 2.71	35.0 ± 3.20	**

* $p < 0.05$.

** $p < 0.01$.

*** $p < 0.001$.

The individual development of each case, boys and girls shown separately, measured as arch widths at canines from age 4 to 6 to 12 is shown graphically in Figs 4a and b, where the mean

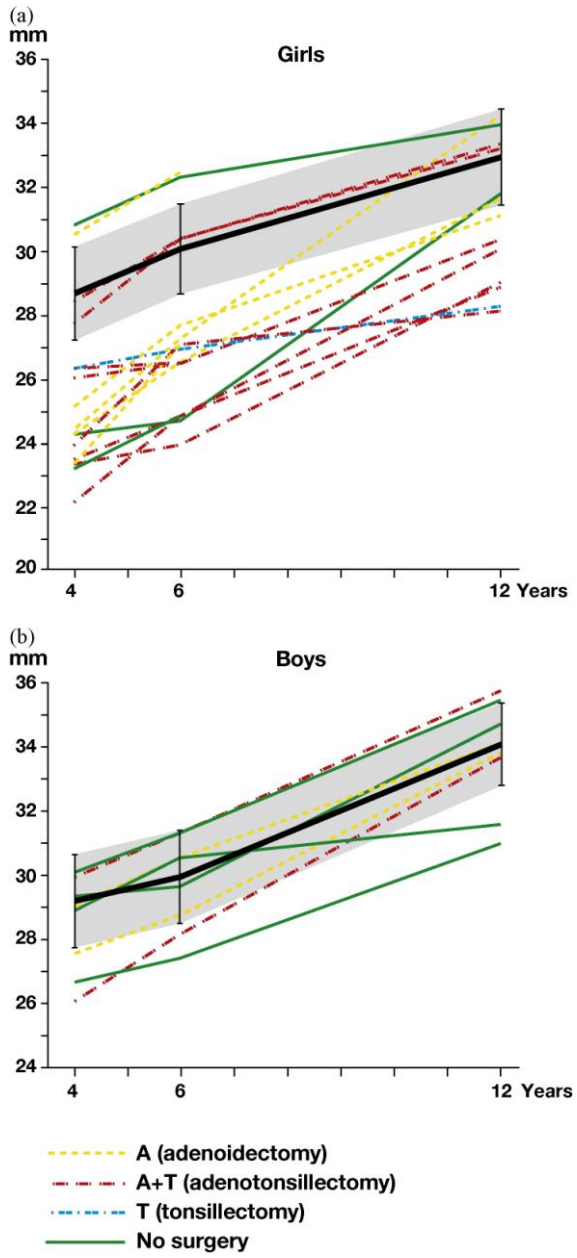

Figure 4 a and b: Individual development of maxillary arch width for each case from age 4 to 6 to 12, as measured at the tip of the canines, in relation to type of surgery or no surgery. Data for girls (a) and boys (b). Mean and standard deviation for corresponding development for controls (girls and boys) is included.

and one SD are also shown for the controls at corresponding ages. 13/17 girls had a more narrow maxilla than the mean value minus one SD among the controls, as did 3/8 boys at age four. Among the operated girls, a catch up in maxillary width between age 4 and 6 is seen for 10/14. The girls with the narrowest maxillas also had a greater gain in width between six and 12 than the controls.

Cephalometry

The seven cases in the present study that were snoring regularly both at age 4 and age 12 had cephalograms taken which could be analyzed from both examinations (see Table 6). All but one of these children had an open mandibular inclination, indicating long face, at age 4. At age 12, four children showed a greater reduction of the mandibular angle than the normative data between 4 and thirteen years. All seven children were breathing orally to some extent at age 12.

Table 6: Mandibular inclination in grades in seven children, snoring every night at ages 4 and 12, and normative data for ages 4 and 13 [17]. Type of surgery or no surgery is denoted.

Child no	**Preop. 4 years**	**Postop. 12 years**			**Normative at 4 years**	**Normative at 13 years**	
		A	A + T	No surg	♂ ♀	♂	♀
1 ♀	38.2	35.1					
2 ♀	34.2	29.5					
3 ♀	40.8	36.3					
4 ♂	28.9		24.9				
5 ♀	40.8		38.5				
6 ♂	37.5			32.4			
7 ♀	34.5			30.4			
Mean SD	36.4 ±4.2				32.5 ±2.9	30.7 ±4.3	29.6 ±4.3

Eight of the cases, who were documented with lateral cephalograms at four years, reported “no snoring” at age 12 and seven reported “snoring every night”. The cephalograms from age 4 from those two groups of children were compared. There were significant differences seen between them and for the children snoring every night at age 12 in comparison to normative data

The size of the combined method error in landmark and measurement-point localisation and measurement was calculated . It did not exceed 0.8 (mm, degrees or distance-ratios).

Discussion

One strength of the present study, which at the same time was a weakness, is the separation in analysis of biometrical data from girls and boys already from age 4. Statistical significance could only be reached among girls because the group of boys among the cases participating from age 4 to 6 to 12 was too small to support statistical significance or to allow reaching valid conclusions when making comparisons to the controls. A recent study of dental arch measurements from age 5, demonstrates gender differences of maxillar width at that age in a normal material [15], which we have been able to observe in our material already at age 4.

In the present study, we have shown that children snoring regularly at age four have a reduced maxillary width compared to those not snoring. This reduced maxillary width is retained throughout childhood independently of adenotonsillar surgery—even in the cases where snoring was reduced. Only a slight catch up of maxillar development was seen after A or A+T. This seems to be in disagreement with earlier investigations [18–20)], that report, especially at younger ages, a strong tendency to normalization of growth after surgery [18]. The difference in results can be due to the longer span of follow-up for the present study.

The cases in the present study came from a cohort of children where all cases were followed longitudinally-even those not operated. Other studies have most often used clinical selection of children and primarily reported the development for the successful cases in comparison to healthy controls [19]. Our findings support the idea that genetics probably have the greatest impact on dental arch morphology.

The greater gain in width that was seen for girls directly after A or A+T can be interpreted as a positive result of the operation despite their still comparatively narrow maxillas. Progressive

narrowing could have been expected if these girls had continued to be snorers and oral breathers.

The cases that were snoring both at age 4 and 12 in many cases showed long face characteristics at both examinations. Cheng et al. (1988) [21] demonstrated that there are intrinsic interrelations of combinations of dental arch and craniofacial characteristics. A recent study of dental casts and facial anatomy in adult subjects confirms these findings, showing that narrow arches are connected to longer faces [22].

In the present study, half of the children who had primarily reduced their snoring after surgery, relapsed by age 12, regardless of the type of operation. At that age, most of them also reported oral breathing, which is a known risk for snoring [23]. The reason for this relapse might have been that the habit of open mouth posture persisted

Half of the cases who were not operated due to parental hesitance had a wider maxilla in comparison to the cases operated. The otorhinolaryngologist might have been less active in persuading these parents to agree to surgery since their child seemed to have normal orthodontic development—this would have been a bias. However, the doctors' risk-evaluations seem to have been correct, since most of these cases reduced their snoring without surgery and at age 12 reported "never" or "sometimes" with respect to oral breathing. The connection between sleep disordered breathing and an open mouth posture seems clear in this study as was the case in earlier studies [11, 12, 24]. However, surgery for snoring does not necessarily result in nasal breathing. Failure to achieve this may be the cause of relapses to snoring and might in some cases explain the lack of positive maxillar development. Postoperative controls should be mandatory and training programs to ensure nasal respiration if nasal breathing is not resumed [25].

Lateral cross-bite and anterior open-bite were seen among the cases as well as in the controls, but were twice as frequent among the cases, both at four and six years of age. These

aberrations can be in part due to prolonged sucking habits [26,27], but since the habit was just as common among the controls as among the cases, the difference cannot be solely due to sucking habits. The malocclusion can also be caused by oral breathing which in accordance to the theory of "functional matrix" by Moss [2], saying that a favorable dental arch growth depends on nasal breathing which makes the tongue a mold for development. The number of cases reporting oral breathing was reduced between ages four and twelve, at which time almost all malocclusions were resolved among both "cured" (not snoring) and "not cured" cases as well as among controls.

When looking at the operated cases divided into "cured" and "not cured" at age 12, the finding that the "cured " (not snoring) had a significant reduced maxillary width compared to the controls is unexpected and not reported earlier. Although the groups are small, this result further supports genetics as the basis for growth and development.

Another reason for snoring in connection to adenoids and tonsils other than reduced maxillary width is overweight—nowadays becoming common [28,29]. However, overweight could not explain failures of surgery in the present group, where all children were of normal weight. Cephalometric data for the not cured children at age twelve, demonstrated long faces both at ages 4 and 12, with a reduction of mandibular inclination even slightly exceeding that of the normative data. Those data (normative) were drawn from a recently published study [17] which reported measurements for ages 10 and 13, but not age 12; the data from age 13 were used in this study even though a slightly wider mandibular angle would be present at 12. The cases which had stopped snoring at age 12 already had slightly different vertical proportions at age 4 indicating more favorable development. This would tend to support the idea that facial type is genetically decided and already observable at an early age [30].

It would be of considerable interest to follow the further development in the present group of children. Guilleminault et al. (1989) [31] have pointed out that children cured by

adenotonsillectomy in childhood can relapse during the teens with both OSAS and negative oro-facial development. We have already seen this development in our study material at age 12.

Conclusion

Children with genetically narrow maxilla have an increased risk of snoring and sleep disordered breathing during the period that there is physiologic hypertrophy of adenoids and tonsils. The maxillar width development in these snoring children is changed very little by adenotonsillar surgery even in the cases where snoring is cured.

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