Sleep Medicine 98 (2022) 98–105

Contents lists available at ScienceDirect

Sleep Medicine

journal homepage: www.elsevier.com/locate/sleep

Prevalence of mouth breathing, with or without nasal obstruction, in children with moderate to severe obstructive sleep apnea

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ARTICLE INFO

Article history:
Received 28 March 2022
Received in revised form
24 June 2022
Accepted 26 June 2022
Available online 30 June 2022

Keywords:
Sleep apnea
Children
Mouth breathing
Nasal obstruction
Rhinometry
Pharyngometry
Cephalometry

ABSTRACT

Background: Mouth breathing (MB) is a symptom of obstructive sleep apnea (OSA) in children, but its diagnosis remains challenging. The main objectives of our study were to evaluate whether parents' and physician's diagnose of MB were concordant and to evaluate the prevalence of nasal obstruction in children with OSA and MB.
Methods: Ninety-three children (median age: 10.6 years, range 3–18) with moderate to severe OSA prospectively underwent otorhinolaryngologist (endoscopy, acoustic rhinometry and pharyngometry allowing calculation of pharyngeal compliance) and orthodontist (clinical exam and cephalometry) assessments together with parental interview (daytime MB: never, sometimes, often, always). MB was also assessed by the otorhinolaryngologist (nasal obstruction on endoscopy) and the orthodontist (incompetent lips or anterior open bite or low tongue position).
Results: Thirty-eight children (41%) were mouth (parental criterion: MB often or always, median age 8.2 years) and 55 nasal (11.4 years, $p = 0.016$) breathers. The agreement of parental and physician diagnosis of MB was slight (orthodontist) to moderate (otorhinolaryngologist). Parental diagnosis of MB was associated with nasal obstruction on acoustic rhinometry and endoscopy (hypertrophy of inferior turbinate, $n = 18$ or adenoids, $n = 15$) and with an adenoid facies (increased Frankfort's mandibular plane angle on cephalometry). Eleven children had MB by habit and were characterized by more severe OSA and higher pharyngeal compliance than mouth breathers with nasal obstruction.
Conclusion: MB diagnosis by parents is acceptable and is mainly related to nasal obstruction. A subset of children had MB by habit associated with worst OSA and increased pharyngeal compliance that could benefit from myofunctional therapy.

1. Introduction

Mouth breathing is a cardinal symptom of obstructive sleep apnea (OSA) in children. For instance, Carroll and colleagues performed a retrospective study of clinical histories and they did not find clinical differences between primary snoring children and those with OSA, except for mouth breathing [1]. Mouth breathing has been further independently associated with the diagnosis of OSA [2] and is a symptom included in questionnaires of OSA such as Pediatric Sleep Questionnaire [3], Selected Features questionnaire [4], Sleep Clinical Record [5], OSA-5 [2] and OSA-18 questionnaires [6]. In these parental questionnaires, "mouth breathing during the day" is the question sometimes referred to as nasal obstruction (OSA-5 and OSA-18). Nasal obstruction has also been independently related to OSA [7], and nasal obstruction in healthy children and adults increases the apnea-hypopnea index (AHI) [8,9].

Adenotonsillectomy is the first-line treatment for pediatric OSA

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https://doi.org/10.1016/j.sleep.2022.06.021
1389-9457/

[10]. Although completely resolved OSA after adenotonsillectomy, some children still show persistence of mouth breathing [11]. It has been hypothesized that many people who mouth-breath have weak oral muscles, which causes them to have their mouth open during the day, mouth breathing is therefore not necessarily related to nasal obstruction defining mouth breathing by habit [12]. Whether mouth breathing is related to nasal obstruction or not deserves to be evaluated from a therapeutic point of view. In the former case, nasal corticosteroids and/or montelukast or nasal surgery could be proposed, while in the latter case, myofunctional therapy should be proposed [13–15]. Nevertheless, the diagnoses of both mouth breathing and nasal obstruction remain challenging issues since agreement for mouth breathing is weak [12] and since there is little consistency between otorhinolaryngologists in their assessment of the degree of nasal obstruction on exam [16].

The main objectives of our prospective, observational, cross-sectional study were to evaluate whether parents' and physician's diagnose of mouth breathing were in agreement and to evaluate the prevalence of nasal obstruction in mouth breathers in a population of children with moderate to severe OSA. The secondary objective was to evaluate whether mouth breathing was associated with modifications of lateral cephalometry parameters. The pertinence of this study is underpinned by the perspective of adapting the therapeutic approach of OSA, especially by guiding ear-nose-throat (ENT) and orthodontic treatments. An additional aim was to evaluate whether mouth breathing was associated with dental and craniomorphologic changes in OSA children.

2. Material and methods

2.1. Design

Primary objectives: Prospective observational study of consecutive otherwise healthy children referred for treatment of their moderate to severe OSA according to ERS recommendations [13]. The primary objectives were to assess the agreement of mouth diagnosis by parents and physicians, and the prevalence of mouth breathing due to nasal anatomical obstruction or unrelated to nasal anatomical obstruction (habit).

Secondary objective: From this prospective cohort, a sample of children was further selected for a case-control experiment to selectively evaluate whether mouth breathing was associated with modifications of lateral cephalometry considering that age, gender and ethnicity may affect the findings [17,18].

2.2. Methods

We conducted the study at a university care center (Robert Debré pediatric hospital, Paris, France) over a 2-year period. This study meets the criteria set by the latest revision of the Declaration of Helsinki and was approved by our local Ethics Committee (PHENOSAS: N° 2018–416); the database of collected data was declared to the French regulatory agency (CNIL). The subjects and their parents were informed of the collection of their prospective data for research purposes, and they could request to be exempted from this study in accordance with French law (non-interventional observational research).

The children (3–18 years) having a moderate to severe OSA (obstructive AHI determined by night-polysomnography ≥5/hour of sleep during a previous hospitalization, as previously described [19,20]) underwent a systematic evaluation on the same day in a multidisciplinary clinic (an orthodontist, an otolaryngologist and a pediatrician specialized in sleep medicine) devoted to the multistep therapeutic approach of childhood OSA, as recommended [13]. The data collected included demographics, ethnicity, z-score of BMI (a nurse measured height, weight and neck circumference), the presence or absence of physician diagnosed asthma, previous adenotonsillectomy and symptoms of snoring and/or sleep-disordered breathing (Brouillette, Spruyt-Gozal and Epworth questionnaires). The sole non-inclusion criterion for this study was the presence of a syndromic disease.

As part of the protocol of the multidisciplinary evaluation, a complete orthodontic assessment was performed including a lateral cephalogram. From the cephalogram, SNA, SNB, ANB, FMA, H-MP distance were calculated (see Fig. 1 for the definitions).

The clinical orthodontic evaluation included the frontal view (vertical relationship, lip relationship, incisor show at rest and on smiling, transverse relationship and symmetry), the profil view (anteroposterior relationship, nasolabial angle and lip protrusion, vertical relationship) and the intraoral examination (dental health, dental arches, static occlusion (incisor relationship, overjet, overbite/underbite, crossbite), functional occlusion). Infantile or visceral swallowing was characterized by a forward movement of the tongue tip.

ENT assessment included endoscopic nasal examination. Clinical evaluation of nasal obstruction was done using Glatzel mirror [21]. A positive simplified Glatzel mirror test defined nasal obstruction when the condensation did not exceed the first line while the child breathed slowly through both nostrils, with no inspiratory or expiratory effort (Fig. 2).

The metal plate was placed horizontally under the nostrils of participants, placing the mirror's zero point under the collumela. Participants were asked to breathe slowly through both nostrils, with no inspiratory or expiratory effort. Condensation not reaching the first line defined a positive mirror test (nasal obstruction) on either right or left side.

Fiber endoscopy was performed with a flexible endoscope after local anesthesia using lidocaine chlorhydrate 10%. Nasal obstruction was defined as obstruction due to adenoid hypertrophy (grade 3 or 4 according to Cassano et al. [22]) or turbinate hypertrophy. Tonsillar hypertrophy was graded according to Brodsky score [23]. The otorhinolaryngologist was blinded to the results of rhinometry.

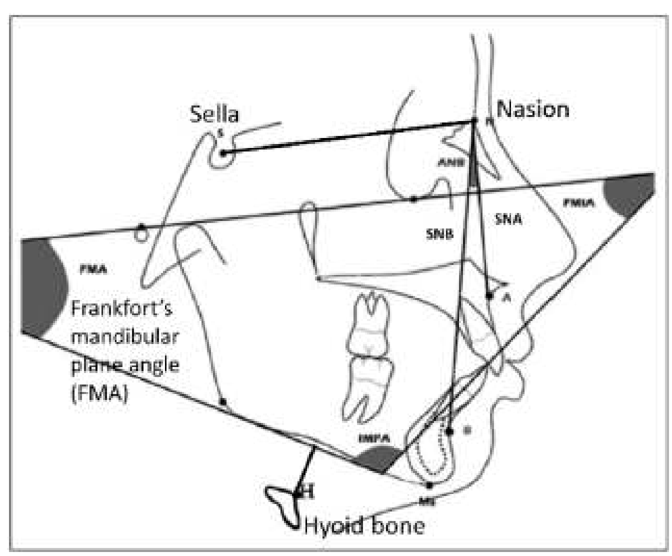

Fig. 1. Measures obtained from the lateral cephalogram.
Measured angles (NV – indicative "normal" value)
SNA is angle between lines S–N and N-A (NV = 80 ± 2°). SNB is angle between lines S–N and N–B (NV = 78 ± 2°). ANB is angle between lines NA and NB. FMA is Frankfort's mandibular plane angle (NV = 25 ± 3°). H-MP is hyoid to mandibular plane distance.

Fig. 2. Simplified Glatzel mirror test.

Patients underwent acoustic rhinometry and pharyngometry testing using an Eccovision 4.50 acoustic rhinometer-pharyngometer (Sleep Group Solutions), as previously described [19,24]. Variables of rhinometry included the minimum cross-sectional area (MCA) and its % predicted value [25], volume of the nasopharynx corrected for height to obtain a normalized parameter and calculated nasal resistance given by the apparatus (from nostril to nasopharynx). Nasal airway resistance was determined for each side of the nose and the total resistance calculated using Ohm's law equation for parallel resistors: $1/R_T = 1/R_r + 1/R_l$, where R_T is the total nasal resistance, R_r = nasal resistance on the right side, R_l = nasal resistance on the left side.

Variables of pharyngometry were the minimal cross-sectional area corresponding to the junction between the oral cavity and oropharynx, the volume of the pharynx (between the oropharyngeal junction and glottis) and the mean pharyngeal area. These measurements were made on patients in sitting and supine positions (after they were in this position for 5 min). An estimated pharyngeal compliance (cm^3/kPa) was then calculated as described previously [19,20,24].

2.3. Definitions of mouth breathing

Mouth breathing was evaluated using three different methods: 1/the notion of daytime mouth breathing (never, sometimes, often, always) was collected from the parents; 2/the otorhinolaryngologist evaluated the presence of daytime mouth breathing related to nasal anatomical obstruction (presence of turbinate and/or adenoid hypertrophy on endoscopy [26]); and 3/the orthodontist evaluated the presence of daytime mouth breathing based on three clinical and cephalogram signs (incompetent lips and/or anterior open bite and/or low tongue position on the mouth floor [27]). Physician's diagnosis of mouth breathing was based on findings of clinical exam only in order to allow a non-biased comparison with the parental criterion. Mouth breathing in our study was defined by the parental criterion (mouth breathing: often or always) since it corresponds to what is asked for in OSA questionnaires.

2.4. Statistical analyses

Sample size calculation: the sample size for a descriptive study of a dichotomous variable (with and without mouth breathing) was based on an expected proportion of mouth breathers of 0.4, a total width of confidence interval of 0.20 and a confidence level of 95%, normal approximation to the binomial calculation gave a sample size of 92 children.

Results were expressed as median [25th–75th percentiles]. Two-group comparisons of continuous variables were performed using the Mann–Whitney *U* test. Categorical variables were compared using the chi-squared test with Yates correction when necessary. A p value < 0.05 was deemed significant. Statistical analyses were performed with StatView 5.0 (SAS Institute) software.

3. Results

3.1. Children with moderate to severe OSA

Ninety-three otherwise healthy children presenting with moderate to severe OSA were included. A moderate agreement for mouth breathing was observed between parental and otorhinolayngologist assessments (% of agreement: 71.0; Cohen's kappa: 0.41), while a slight agreement was observed between parental and orthodontist assessments (% of agreement: 58.1; Cohen's kappa: 0.17).

Among these 93 children, 38 (41%, 95% confidence interval [CI]: 31–52%) were classified as mouth breathers and 55 as nasal breathers (based on parental criterion). Table 1 describes their respective characteristics. Mouth breathers were younger, had more OSA symptoms and had nasal obstruction: mouth breathers as compared to nasal breathers had more frequently positive Glatzel mirror tests and inferior turbinate hypertrophy; the former also had reduced valve area (% predicted), reduced corrected naso-pharyngeal volume and logically increased nasal resistance. Furthermore, mouth breathers depicted incompetent lips and infantile swallowing. Their measures obtained from lateral cephalogram were not significantly different with the exception of a higher FMA in mouth breathers than nasal breathers.

Ethnicity: C is Caucasian, B is African, A is Asian and M is mixed; BMI is body mass index; OAHI is obstructive apnea-hypopnea index; ODI is oxygenation desaturation index; SpO_2 is arterial saturation; lateral cephalogram measurements are described in Fig. 1.

Infantile (visceral) shallowing is that which exists at birth and is characterized by a forward movement of the tongue tip. This atypical swallowing is a myofunctional problem consisting of an altered tongue position during the act of swallowing [28].

ND denotes not done due to parental definition criterion of mouth breathing. Significant results are in bold.

3.2. Different categories of mouth breathers

We first evaluated the agreement between rhinometry and clinical assessment by the otorhinolaryngologist (Glatzel mirror and endoscopy results). First, a positive simplified Glatzel mirror test was associated with increased nasal resistance (right side, p = 0.011; left side, p = 0.008). Inferior turbinate hypertrophy (from mild to severe) was associated with decreased MCA (p = 0.032) while adenoid hypertrophy was associated with decreased corrected naso-pharyngeal volume (p = 0.011).

Then, nasal obstruction was defined based on otorhinolaryngologist findings (presence of turbinate hypertrophy or/and adenoid hypertrophy). Mouth breathers with nasal obstruction represented 27/38 mouth breathers (71%, 95% CI: 54–85%) while mouth breathers by habit (without nasal anatomical obstruction) were the others (n = 11).

We further evaluated whether nasal resistance measurement could predict nasal obstruction (based on endoscopy findings). A nasal resistance >3.93 $cmH_2O/L/s$ had a sensitivity of 0.80 (95% CI: 0.66–0.90) and a specificity of 0.60 (95% CI: 0.42–0.76) to predict

Table 1
Characteristics of the 93 participants according to breathing route (parental criterion).

Characteristics	Mouth breathing N = 38	Nasal breathing N = 55	P value
Gender, Female/male	14/24	20/35	>0.999
Ethnicity, C/B/A/M	17/16/4/1	28/21/2/4	0.421
Age, years	8.2 [6.3; 12.1]	11.4 [8.9; 13.4]	**0.016**
Height, cm	137.0 [121.5; 161.0]	152.0 [139; 162.7]	0.060
Weight, kg	34.5 [21.9; 76.0]	56.0 [35.6; 92.5]	0.100
z-score BMI	1.56 [-0.08; 2.44]	2.00 [0.67; 2.53]	0.556
Neck circumference, cm	29.5 [26.7; 36.0]	33.0 [28.5; 38.0]	0.137
Asthma	8	4	0.064
Previous adenotonsillectomy	18	15	0.052
Brouillette questionnaire	3.97 [2.55; 3.97]	1.14 [-1.35; 2.91]	**0.001**
Spruyt-Gozal questionnaire	3.11 [2.50; 3.62]	2.25 [1.11; 2.98]	**0.002**
Epworth questionnaire	8 [3; 12]	6 [3; 10]	0.349
Mouth breathing, never/sometimes/often/always	0/0/12/26	38/17/0/0	ND
Apneas reported by parents, nev/som/oft/alw[a]	4/5/9/20	23/9/13/10	**0.001**
Polysomnography			
OAHI/hour of sleep	10.4 [6.5; 19.4]	8.6 [6.4; 20.3]	>0.999
ODI/hour	9.1 [5.1; 16.6]	6.7 [4.4; 16.8]	0.417
SpO_2 nadir, %	86 [85; 90]	88 [83; 91]	0.467
Otorhinolaryngologist, clinical exam			
Positive Glatzel mirror test, right	8	0	**<0.001**
Positive Glatzel mirror test, left	7	0	**<0.001**
Brodsky grade, 1–2/3-4	20/18	30/25	0.856
Hypertrophy of inferior turbinate, n	18	5	**<0.001**
Hypertrophy of adenoids, n	15	12	0.103
Mouth breathing criterion, n	27	16	**0.001**
Acoustic rhinometry			
Right valve area, cm^2	0.28 [0.18; 0.34]	0.37 [0.27; 0.45]	**0.001**
Left valve area, cm^2	0.30 [0.22; 0.37]	0.37 [0.23; 0.47]	**0.043**
Mean valve area, cm^2	0.27 [0.22; 0.37]	0.37 [0.27; 0.47]	**0.001**
Valve area, % predicted	65 [50; 76]	77 [57; 99]	**0.010**
Right naso-pharyngeal volume, cm^3	5.2 [3.6; 9.1]	7.5 [6.2; 9.0]	**0.019**
Left naso-pharyngeal volume, cm^3	6.6 [3.8; 8.0]	7.7 [5.6; 10.6]	**0.038**
Corrected naso-pharyngeal volume	0.33 [0.22; 0.49]	0.42 [0.29; 0.49]	**0.038**
Right nasal resistance, $cmH_2O/L/s$	11.0 [4.6; 17.1]	5.3 [4.0; 8.7]	**0.007**
Left nasal resistance, $cmH_2O/L/s$	7.6 [4.5; 11.5]	4.8 [3.4; 11.6]	0.113
Nasal Resistance, $cmH_2O/L/s$	4.1 [2.4; 6.4]	2.6 [1.8; 3.8]	**0.007**
Acoustic pharyngometry			
Sitting oropharyngeal junction area, cm^2	1.22 [0.94; 1.45]	1.38 [0.99; 1.58]	0.124
Sitting pharyngeal volume, cm^3	17.7 [13.6; 23.5]	18.6 [14.5; 25.2]	0.527
Pharyngeal compliance, cm^3/kPa	10.90 [1.68; 16.77]	9.57 [3.04; 19.65]	0.881
Orthodontist, clinical exam and cephalometry			
Asymmetry, n	4	1	0.155
Facial height, decreased/normal/increased	1/27/10	1/48/6	0.142
Profil view, cis/ortho/trans	4/19/13	3/33/19	0.572
Nasolabial angle, closed/normal/open	6/24/5	11/41/3	0.354
Chin, protrusion/normal/retrusion	2/31/5	0/43/12	0.146
Anterior open bite, n	9	5	**0.042**
Incompetent lip seal, n	15	8	**0.006**
Low and forward tongue position, n	20	21	0.128
Mouth breathing criterion, n	24	25	0.139
Infantile swallowing, n	27	24	**0.002**
Dental class, I/II/III[b]	15/3/9	24/3/11	0.807
Lateral cephalogram, n	27	38	
SNA	83 [81; 87]	84 [82; 87]	0.367
SNB	79 [77; 83]	80 [77; 84]	0.603
ANB	4.0 [2.0; 5.0]	4.0 [2.0; 6.0]	0.723
FMA	28.0 [23.2; 32.2]	23.0 [19.5; 27.0]	**0.003**
H-MP distance, mm	12.5 [11.0; 18.2]	13.2 [9.8; 17.0]	0.754

[a] nev/som/oft/alw denotes never/sometimes/often/always.
[b] Dental class occlusion: I is normal occlusion, class II is where the lower first molar is posterior (or more towards the back of the mouth) than the upper first molar and class III is where the lower first molar is anterior (or more towards the front of the mouth) than the upper first molar.

nasal obstruction (area under the curve: 0.71, 95% CI: 0.60–0.83), p = 0.0004 (Youden method).

Table 2 shows the characteristics of the two subgroups of mouth breathers. Mouth breathers by habit (without nasal anatomical obstruction) had a lower nasal resistance and more importantly had a more severe OSA associated with a higher pharyngeal compliance than mouth breathers with nasal obstruction.

Ethnicity: C is Caucasian, B is African, A is Asian and M is mixed; BMI is body mass index; OAHI is obstructive apnea-hypopnea index; lateral cephalogram measurements are described in Fig. 1.

ND denotes not done due to ENT definition criteria of mouth breathing. Significant results are in bold.

3.3. Secondary objective of the study: case-control study

We hypothesized that the absence of significant differences (with the exception of FMA) for cephalogram data could have been related to the difference of age of the two groups. Moreover,

Table 2
Characteristics of the 38 mouth-breathers according to the presence of nasal obstruction.

Characteristics	Nasal anatomical obstruction N = 27	No nasal anatomical obstruction, habit N = 11	P value
Gender, Female/male	11/16	3/8	0.488
Ethnicity, C/B/A/M	12/12/2/1	5/4/2/0	0.707
Age, years	7.7 [5.8; 11.7]	11.7 [7.6; 12.8]	0.079
z-score BMI	0.38 [-0.26; 2.20]	2.00 [1.61; 2.55]	0.064
Neck circumference, cm	28.0 [26.0; 34.8]	35.5 [30.2; 37.5]	**0.047**
Brouillette	3.97 [2.55; 3.97]	2.56 [-0.11; 3.97]	0.279
Spruyt-Gozal	3.12 [2.64; 3.50]	2.75 [4.41; 6.62]	0.747
Epworth	8 [4; 12]	8 [3; 10]	0.448
Apneas reported by parents 0/1/2/3	2/5/7/13	2/0/2/7	0.327
Asthma	4	4	0.195
Previous adenotonsillectomy	10	8	0.074
OAHI/hour of sleep	7.6 [6.3; 10.1]	16.1 [8.4; 26.0]	**0.047**
Otorhinolaryngologist, clinical exam			
Positive Glatzel mirror test, right	8	0	ND
Positive Glatzel mirror test, left	7	0	ND
Brodsky grade, 1–2/3-4	14/13	6/5	>0.999
Hypertrophy of inferior turbinate, n	18	0	ND
Hypertrophy of adenoids, n	15	0	ND
Acoustic rhinometry			
Mean valve area, cm^2	0.25 [0.19; 0.28]	0.37 [0.27; 0.47]	**0.008**
Valve area, % predicted	60 [40; 71]	77 [57; 99]	0.082
Corrected naso-pharyngeal volume	0.23 [0.20; 0.47]	0.42 [0.29; 0.49]	0.136
Nasal Resistance, $cmH_2O/L/s$	5.3 [3.2; 7.6]	2.6 [1.8; 3.8]	**0.028**
Acoustic pharyngometry			
Sitting oropharyngeal junction area, cm^2	1.11 [0.93; 1.32]	1.44 [1.03; 1.53]	0.219
Sitting pharyngeal volume, cm^3	16.3 [13.3; 22.0]	22.0 [17.8; 28.9]	0.073
Pharyngeal compliance, cm^3/kPa	8.51 [1.37; 13.16]	17.32 [9.01; 21.29]	**0.028**

averaging cephalometric data from boys and girls of different ethnicities could also have biased our results. Thus, we performed an additional experiment that consisted in a case-control study. We selected mouth breathing children with nasal obstruction (n = 27) and nasal breathers defined by the absence of mouth breathing (n = 55). Then, cases (mouth breathers) and controls (nasal breathers) were matched for gender, ethnicity and age (±0.5 year) giving 13 pairs of children having had cephalometry. The characteristics of the two matched groups of children are given in Table 3.

Ethnicity: C is Caucasian, B is African, A is Asian and M is mixed; BMI is body mass index; OAHI is obstructive apnea-hypopnea index; lateral cephalogram measurements are described in Fig. 1. ND denotes not done due to matching criteria. Significant results are in bold.

We confirm in this subgroup of children that FMA is increased in mouth breathers and we further evidence an increase in hyoid bone to mandibular plane distance as compared to nasal breathers.

4. Discussion

The main results of our prospective observational study were first to show that the agreement of parental and physician diagnosis of daytime mouth breathing was slight to moderate only. Nevertheless, parental diagnosis of mouth breathing was associated with some degree of nasal obstruction on acoustic rhinometry and with an adenoid facies (characterized by incompetent lip seal, open anterior bite and increased FMA). In our study, ENT exam agreed quite well with the results of rhinometry, which allowed us to determine that mouth breathing was associated with nasal obstruction in the majority of children. Children with mouth breathing by habit were characterized by more severe OSA associated with higher pharyngeal compliance than mouth breathers with nasal obstruction.

4.1. The diagnosis of mouth breathing

Recognition of mouth breathing in children is challenging [29]. Detecting airflow by carbon dioxide sensor can discriminate breathing mode, but the degree of nasal resistance and subjective symptoms of mouth breathing do not accurately predict breathing mode [30]. In this latter study, nasal resistance was significantly greater for the mouth breathers than for the nasal breathers,

Table 3
Characteristics of the 13 pairs of participants according to breathing route.

Characteristics	Mouth breathing N = 13	Nasal breathing N = 13	P value
Gender, Female/male	5/8	5/8	ND
Ethnicity, C/B/A/M	7/6/0/0	7/6/0/0	ND
Age, years	9.6 [7.3; 13.4]	10.1 [7.3; 13.9]	0.898
z-score BMI	1.93 [0.26; 2.70]	2.15 [-0.76; 2.63]	0.857
Neck circumference, cm	34.0 [26.4; 37.4]	33.3 [27.4; 38.0]	0.979
OAHI/hour of sleep	12.4 [8.4; 24.8]	13.7 [7.0; 21.8]	0.786
Lateral cephalogram			
SNA	83 [81; 87]	84 [80; 85]	0.643
SNB	79 [77; 81]	78 [74; 81]	0.354
ANB	4.0 [2.0; 6.2]	5.0 [1.7; 6.5]	0.718
FMA	30.0 [27.7; 35.2]	26.0 [22.2; 27.5]	**0.011**
H-MP distance, mm	15.0 [14.4; 21.7]	11.0 [8.9; 16.5]	**0.029**

suggesting that nasal resistance may help for mouth breathing diagnosis. We further confirm that the sensitivity of nasal resistance measurement is 80% in our hands. Nevertheless, exclusive nasal breathing has been found over a wide range of nasal resistances, many subjects with open lips [31]. Finally, some children are mouth breathers by habit, without any nasal obstruction [29]. Thus, both clinical (absence of lip seal) and functional (nasal resistance) criteria are imperfect.

We show that the agreement of parental and physician diagnosis of daytime mouth breathing was slight to moderate only. The agreement with the otorhinolaryngologist was moderate, which is explained by the fact that the physician diagnosis relied on the exclusive nasal obstruction criteria while parents can diagnose children with mouth breathing by habit. The agreement with the orthodontist was slight only. The three clinical criteria chosen for orthodontic diagnosis are probably more specific than sensitive. Along this line, it has been shown that there is no standardization of recognition of mouth breathing among orthodontists [12]. Lack of lip seal and presence of anterior open bite were among the most frequent orthodontic criteria [12]. We added the low tongue position criterion that is linked to high arched palate, which had a higher odds ratio than reduced hard palate width to predict mouth breathing [27]. Using standardized criteria including breathing tests (Glatzel mirror test, water retention test and lip seal test), this study showed that absence of lip seal was observed in 36% of mouth breathers (15/38, 39% in our study) while its presence was observed in 97% of nose breathers [12]. Anterior open bite was found among 23% of their mouth breathers, while it was found in 9/38 (24%) children in our series. Narrow palate was found in 54% of mouth breathers in their study, while low (and forward) position of the tongue, which is supposed to be responsible for narrow palate, was found in 20/38 (53%) children in our study. Overall, our results are in agreement with the data of Pacheco and colleagues [12]. One may therefore state that parental diagnosis of mouth breathing seems reasonable since it is associated with documented nasal obstruction, explaining its consistent independent link with OSA diagnosis. The percentage of mouth breathers in our study (41%, 95% CI: 31–52%) is consistent with that previously evidenced by the study of Villa and colleagues (52%) [5].

4.2. The diagnosis of nasal obstruction and determination of children with mouth breathing by habit

A gold standard objective measure of nasal airway obstruction does not currently exist, so patient-reported measures are commonly used, which is problematic in young children. Methods of measuring nasal obstruction outcomes include both objective anatomic and physiologic measurements. Anatomic measurements include acoustic rhinometry, imaging studies, and clinician-derived examination findings. Physiologic measures include rhinomanometry, nasal peak inspiratory flow, and computational fluid dynamics. Several studies attempted to draw correlation of these outcome measures; however, few show strong correlation [32]. In our hands clinical and acoustic rhinometry evaluations of nasal obstruction were in good agreement, as previously described in children [33,34]. We did not record allergic rhinitis that has been associated with nasal obstruction nor treatments that may influence nasal obstruction, which is a limitation. Nevertheless, we were confident with the determination of the group of children with mouth breathing by habit, i.e. without anatomical nasal obstruction. Interestingly, these children had more severe OSA that could be related to a higher increase in pharyngeal compliance. Many people who mouth-breath have weak oral muscles in spite of higher EMG activity of muscle from the lips that has been demonstrated in subjects without competent lips implying a higher muscular effort due to the requirement of lip sealing [35]. Therefore, "habit" can possibly be from other neuromuscular issues. Our results may further suggest that oral muscle fatigue is associated with pharyngeal muscle fatigue causing increased pharyngeal compliance. A decrease in oropharyngeal cross-sectional area has previously been demonstrated in supine position despite an increase in genioglossus EMG activity [36]. Thus, subjects without lip sealing will logically have increased pharyngeal compliance. We already suggested that "normal" or decreased pharyngeal compliance before adenotonsillectomy may be related to pharyngeal muscle activation that resolved after surgery, suggesting a protective mechanism against apnea. On the opposite, increased preoperative compliance (relaxation) could be due to muscle fatigue [24]. Thus, the positive relationship between increased compliance and AHI [19] would be explained by pharyngeal muscle fatigue, which seems further increased in mouth breathers by habit. Whether or not mouth breathing by habit occurs after a period of nasal obstruction that resolves remains open to debate. Nevertheless, we observed a trend for more previous adenotonsillectomy in this group (Table 2).

4.3. Dental and craniomorphologic changes

A meta-analysis of lateral cephalometric data of mouth breathers versus nasal breathers has been performed [37]. In this systematic review, mouth breathers demonstrated an increased mandibular plane angle, total and lower anterior facial height and decreased posterior facial height. The increase in FMA in mouth breathers is consistent with the increase in total and lower anterior facial height. This increase, together with incompetent lip seal, is a characteristic of the adenoid facies, which remained to be demonstrated in children with moderate to severe OSA. Indeed, Villa and colleagues did not find an increased prevalence of adenoid phenotype in OSA children as compared to children with primary snoring [5]. We further show that after adequate control for age, gender and ethnicity, an inferiorly positioned hyoid bone was evidenced in mouth breathers. Matching was mandatory since age and gender effects on hyoid bone position have been demonstrated [38]. An inferior position of hyoid bone in children with OSA as compared to children without OSA has already been demonstrated [39]. Kohno and colleagues have demonstrated a reduction of H-MP distance under anesthesia in OSA adults [40]. Caudally located hyoid bone in patients with OSA is a result of interactions between the infra-hyoid muscle activation, compensatory displacement of the excessive soft tissue outside the enclosure and tracheal traction by lung inflation [40]. We further suggest that an increased inferior position is associated with mouth breathing as compared to nasal breathing that could be related to compensatory muscle activation. Along this line, it has been demonstrated that men with a lower hyoid bone show greater genioglossus reflexive response to partial oropharyngeal obstruction [41]. This muscle activation is not incompatible with fatigue of other muscles since hyoid muscular activity was not modified by the presence or absence of lip competence [35]. Along this line, it has been shown that hyoid muscles may compensate for rapid fatigue of the tongue muscle to maintain tongue pressure by changing their activity pattern during tongue pressure generation [42].

4.4. Clinical consequences

Mouth breathing diagnosis and its cause (presence or absence of anatomical nasal obstruction) would be systematically assessed in children suffering from moderate to severe OSA since it will orientate the stepwize treatment approach: the presence of tonsil hypertrophy and/or turbinate hypertrophy will guide the surgery, while children with normal upper airways and mouth breathing by habit should be prioritized for myofunctional therapy. When there are doubts about the degree of nasal obstruction related to adenoid or turbinate hypertrophy, which is probably frequent [32], acoustic rhinometry may help to demonstrate functional consequences, i.e. decreased nasal valve % predicted or decreased corrected naso-pharyngeal volume.

5. Conclusion

Mouth breathing diagnosis by parents is acceptable and is mainly related to nasal obstruction but in a subset of children mouth breathing is due to habit. This latter phenotype is associated with worst obstructive sleep apnea and increased pharyngeal compliance.

CRediT authorship contribution statement

Plamen Bokov: Conceptualization, Methodology, Investigation, Writing – original draft, Supervision. **Jacques Dahan:** Validation, Investigation, Writing – review & editing. **Imène Boujemla:** Validation, Investigation, Writing – review & editing. **Benjamin Dudoignon:** Validation, Investigation, Writing – review & editing. **Charles-Victor André:** Validation, Investigation, Writing – review & editing. **Selim Bennaceur:** Validation, Investigation, Writing – review & editing. **Natacha Teissier:** Validation, Investigation, Writing – review & editing. **Christophe Delclaux:** Conceptualization, Methodology, Formal analysis, Writing – original draft, Supervision, Project administration.

Declaration of competing interest

None.

This study was not funded.

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