Original Article

Teenage sleep-disordered breathing: Recurrence of syndrome

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ABSTRACT

Objectives: The study aims to better understand the reappearance of sleep apnoea in adolescents considered cured of obstructive sleep apnoea (OSA) following adenotonsillectomy and orthodontic treatment.
Study design: The study employs a retrospective analysis of 29 adolescents (nine girls and 20 boys) with OSA previously treated with adenotonsillectomy and orthodontia at a mean age of 7.5 years. During follow-up at 11 and 14 years of age, patients were clinically evaluated, filled the Pediatric Sleep Questionnaire (PSQ) and had systematic cephalometric X-rays performed by orthodontists. Polysomnographic (PSG) data were compared at the time of OSA diagnosis, following surgical and orthodontic treatment and during pubertal follow-up evaluation.
Results: Following the diagnosis of OSA and treatment with adenotonsillectomy and rapid maxillary expansion (Apnea–Hypopnea Index (AHI) 0.4 ± 0.4), children were re-evaluated at a mean age of 11 years. During follow-up at 14 years, all children had normal body mass indices (BMIs). Teenagers were subdivided into two groups based on complaints: Nine asymptomatic subjects (seven girls and two boys) and 20 subjects with decline in school performance, presence of fatigue, indicators of sleep-phase delays and, less frequently, specific symptoms of daytime sleepiness and snoring. Presence of mouth breathing, abnormal AHI and RDI and significant reduction of posterior airway space (PAS) was demonstrated during repeat polysomnography and cephalometry. Compared to cephalometry obtained at a mean of 11 years of age, there was a significant reduction of PAS of 2.3 ± 0.4 mm at a mean age of 14 years.
Conclusion: Previously suggested recurrence of OSA during teenage years has again been demonstrated in this small group of subjects. Prospective investigations are needed to establish frequency of risk, especially in non-orthodontically treated children.

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1. Introduction

Obstructive sleep apnoea syndrome (OSAS) in adults is an important public health problem that can lead to behavioural and cognitive dysfunction, cardiovascular risk and negatively impact quality of life. Adult OSAS is related to a dyscoordination of upper airway dilator muscles and inspiratory muscles during sleep. This dyscoordination can be explained in part by local neuropathy in sleep apnoea [1,2]. Nasal continuous positive airway pressure (CPAP) treatment appears to be the only treatment capable of completely controlling the problem in these cases. However, many adult patients fail to achieve complete control of their OSAS due to non-compliance with CPAP, particularly in older teenagers and young adults.

Significant efforts have been made to recognise the presence of abnormal breathing during sleep at an early age in hopes of preventing the progression of the complete syndrome. OSA can be recognised and treated during childhood. Nevertheless, even with early childhood intervention, long-term treatment may be required. In some cases, children deemed cured of OSA after adenotonsillectomy during prepubertal years relapse in their late teens [3,4]. With this in mind, we have made efforts to better understand the reappearance of sleep apnoea during teenage years. Normally, once a child's symptoms of sleep-disordered breathing (SDB) resolve in the setting of a normal postoperative polysomnogram (PSG), no long-term follow-up testing is performed. There is little information available regarding the course of breathing beyond the immediate postoperative period. In fact, the only published series of children with long-term follow up after OSA treatment involves orthodontic treatment of 10 prepubertal children with 3-year follow-up [5].

Parents of pubertal teenagers reported return of symptoms despite treatment with adenotonsillectomy and/or orthodontia at an
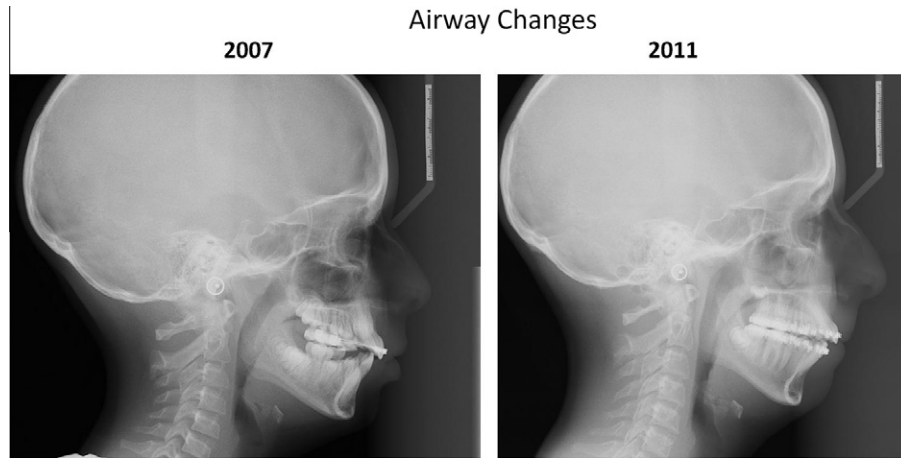

Airway Changes

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Fig. 1. Subjects involved in the study. Graph of the retrospective chart investigation to find the candidates for the retrospective study.

earlier age. We queried how many of such cases we could identify and what interval history could be documented. A systematic search involving three different sleep clinics revealed that such an investigation was difficult due to absence of systematic follow-up information in many cases (see Fig. 1). This report presents limited findings obtained during a retrospective study of children diagnosed with SDB and treated with adenotonsillectomy and orthodontics during their prepubertal years, then later evaluated and treated by orthodontists for dental and/or facial aesthetic indications. The primary goal of this retrospective report was to understand the characteristics of children diagnosed and successfully treated for OSA following adenotonsillectomy and orthodontia during prepubertal years who later presented for orthodontics with recurrence of symptoms and/or signs associated with SDB after the onset of puberty.

2. Methods

2.1. Sources of clinical information

Due to long-term collaborations between three sleep clinics, private and university orthodontic clinics and a craniofacial department, specialists in these clinics have been keenly aware of the presence of SDB and OSA in young individuals. Therefore, as part of a routine clinical evaluation of pediatric patients, the Pediatric Sleep Questionnaire (PSQ) or its validated translation in Chinese [6,7] is given to all children who present for orthodontic evaluation in both the private orthodontic clinic and university-based orthodontic clinics. If the questionnaire suggests a sleep problem, the child is referred to pediatric sleep clinics based upon insurance criteria. Information obtained from these sleep clinic referrals is then sent back to the orthodontic practice.

We used the following inclusion criteria: (1) diagnosis of OSA based on the presence of clinical symptoms investigated by a paediatric sleep medicine specialist and results of an in-laboratory PSG, (2) referral for treatment for OSA as outlined by an expert specialised team (in the the locations, the diagnostic and treatment recommendation involved the combined expertise of otolaryngologist, orthodontist and sleep medicine specialist), (3) having had post-treatment follow-up with clinical evaluation by the clinical team and PSG and (4) having been considered as exempt from SDB at the end of treatment based on parent reports, clinical evaluation and findings and nocturnal PSG. Syndromic and obese children with OSA were excluded from this review. Obesity was an uncommon observation at the time in the three practices.

Unidentified information collected in the universities and private practices was reviewed, including results of the PSQ, results of the orthodontic and sleep medicine evaluations at each available visit, cephalometric radiograph readings and PSG test results.

2.1.1. Chart review

An initial review showed that there was no systematic long-term follow-up schedule through sleep medicine services, as these services were referral clinics. There was also no long-term follow-up in the otolaryngology clinics. The only long-term clinical follow-up available was through the orthodontic clinics (see Fig. 1).

Chart records at the orthodontic–craniofacial clinics were reviewed to identify pubertal individuals (age 12–16 years) who were diagnosed with SDB and appropriately treated with adenotonsillectomy usually followed by rapid-maxillary-expansion (RME) treatment during their prepubertal years [5,8]. There were a total of 35 pubertal individuals seen in an orthodontic clinic who met this requirement. All unidentified information on these individuals was included for analysis.

There were Institutional Review Board (IRB) approvals for evaluation of the previously obtained clinical and PSG data rendered anonymous for research purposes.

2.2. Data collection

In reviewing the retrospective data for potential subjects, we developed three sets of data, described below. To be presented in this retrospective review, subjects must have data available at three different times:

(1) Time of initial diagnosis of SDB as a young child with objective demonstration of resolution of the SDB problem;
(2) Intermediate evaluation at some point after initial diagnosis and treatment period at a mean age of 11 years; this age was selected based on the relationship between age and facial growth;
(3) Facial presentation and breathing evaluation as a pubertal teenager.

As shown in Fig. 1, despite a large review of children, a low number of cases could be extracted. Orthodontic clinics had follow-up for dental, facial aesthetics or routine yearly post-procedural follow-up for rapid maxillary or bimaxillary expansion [8]. The extracted data were as follows:

2.2.2. +Data set 'Prepubertal and young age – (time 1-T1-)'

The data included: initial clinical complaints and symptoms, pediatric, otolaryngological and sleep evaluations and polysomnographic results. Surgical intervention follow up information and follow-up polysomnography were also collected. Age at time of referral to the sleep clinic, gender, height and weight (to derive BMI), reason for the referral, clinical upper airway evaluation, results of initial cephalometric X-ray at time of referral, results of the PSQ, results of sleep clinic evaluation and sleep tests were retrieved.

2.2.3. +Data set 'older-prepubertal children – (time2-T2-)': (mean age 11 years)

Age, BMI, clinical complaints, clinical examination, results of PSQ and cephalometric data were also retrieved.

2.2.4. Data set 'pubertal teenagers – (time 3-T3-)'

These data included results of orthodontic and sleep clinics' re-evaluation covering PSQ results, new clinical evaluation of upper airway anatomy, cephalometric X-rays results and results of a new PSG or polygraphy.
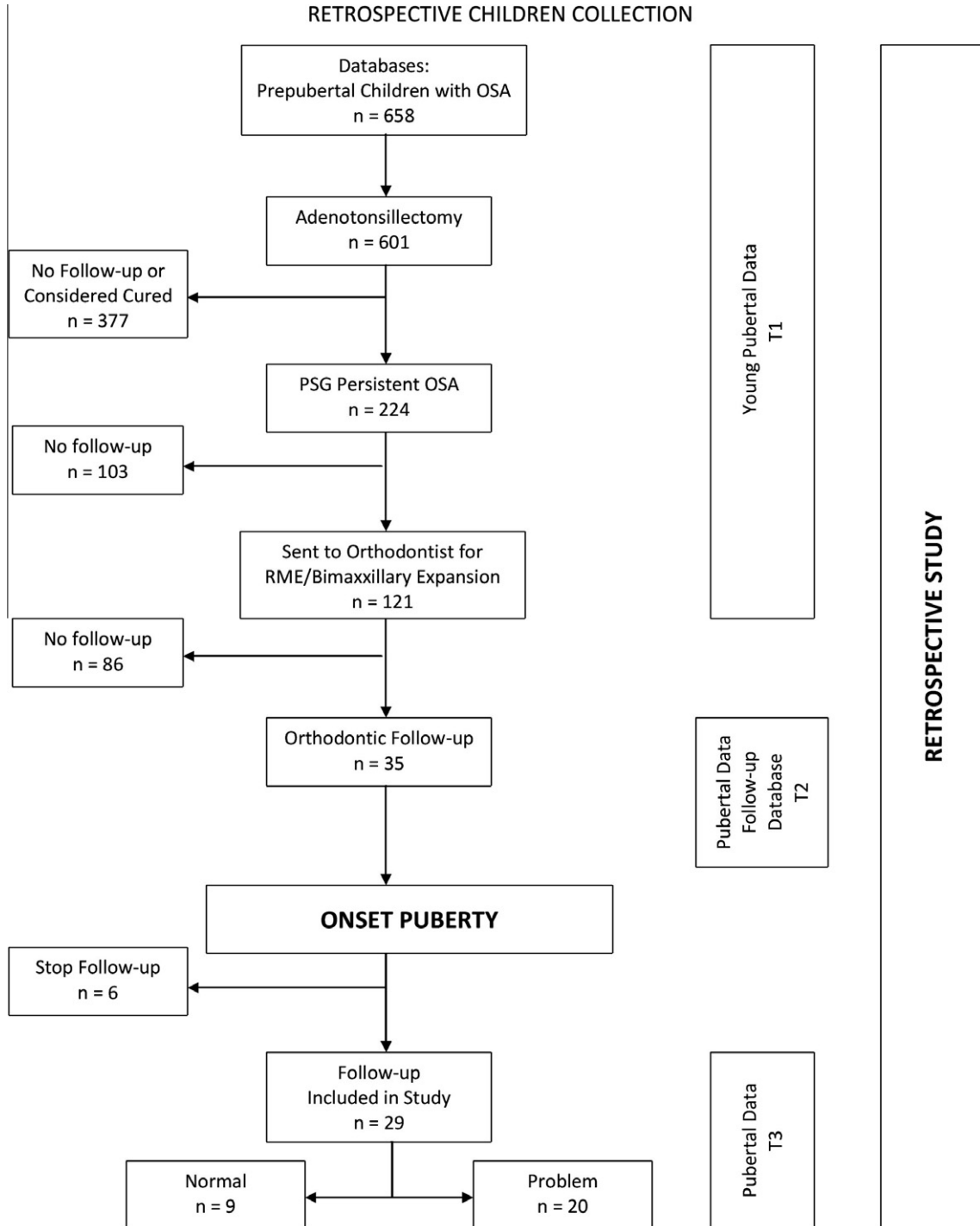

Fig. 2. Cephalometrics at 9.8 and 13.8 years in a boy. Example of the changes noted in the posterior-airway size between pre-pubertal and pubertal.

The clinical new upper airway evaluation included presence of mouth breathing at rest when distracted or performing intellectual tasks; indication of narrow maxilla or mandible, evidence of bruxism. Particularly if partial tonsillectomy had been performed, there was a grading of tonsils based on the Friedman et al. scale [9], grading of upper airway space using the Friedman-modified Mallampati scale with 4 scores [10] and determination of presence of septum deviation and of nasal inferior turbinate hypertrophy based on the mention of "abnormally enlarged" or "hypertrophic" in the chart.

Once all data were collected, results were placed in a spreadsheet for analyses.

To unify PSG information and avoid inter-scorer variability, all polygraphic recordings performed during the pubertal period were obtained and rescored anonymously by one individual for the purpose of the study. As recordings were obtained on different computerised sleep systems, PSG data collected on systems other than Sandman™ were transformed into European Data Format (EDF). All cephalometric X-rays were also reviewed blindly by one individual for consistency of measurements.

During the review, we found that we had 35 cases that had been seen at T1 and T2, but only 29 individuals had data collected at the three required time-points. These 29 subjects represent the 'reviewed group' (Fig. 1).

2.3. Analyses

Sleep scoring was performed using the international manual from Rechtschaffen and Kales [11]. This scoring manual had been used previously to score PSG at any age. It was based on a sleep montage that always included: 4-EEG (electroencephalography) channels, one-chin electromyograph (EMG) and two-leg EMGs; two electrooculography (EOG) leads, one electrocardiograph (ECG) derivation and a position sensor.

Respiratory variables had been measured using similar technology overtime and location with systematic monitoring of the nasal cannula-pressure transducer, mouth thermistor thoracic and abdominal belts, either piezo-electric (mostly in the recordings obtained at a young prepubertal age) or inductive plethysmography (older prepubertal and in all cases during pubertal recordings), diaphragmatic-intercostal EMG, neck microphone and pulse oximetry from which oxygen saturation and finger plethysmography tracings were derived. Depending on age and location, end-tidal or transcutaneous CO_2 was also monitored. Respiratory variables had been scored using the international recommendations defining apnoea and hypopnoea similar to those outlined in the AASM "recommended" criteria [12,13] and the AHI was calculated. A respiratory-disturbance index was also calculated scoring respiratory event related arousals, RERAs, defined as events in which there is a drop in the amplitude of the nasal cannula tracing curve by at least 30% compared to prior normal tracing. Presence of snoring or heavy breathing and increase in snoring signal from beginning to the end of an evening that is associated with the presence of an increase in respiratory efforts was seen on the thoracic and abdominal tracing curves, with termination by an EEG arousal and return to normal amplitude of the nasal cannula curve [14]. EEG arousal was based on the international recommendation [15]. The duration of all events (apnoea, hypopnoea and RERA) was a minimum of 10 s. The apnoeas were further subdivided into obstructive, mixed or central sub-types following the international definitions. Mouth breathing during sleep was documented by the mouth thermistor. The time spent with 'flow limitation' based on the curve obtained from the nasal cannula was calculated at T3. The definition of flow limitation was based on the Hosselet et al. report [16] and a percentage of time spent presenting flow limitation was calculated following prior report [17].

Such montages and monitoring were used in all reported cases except for nine pubertal teenagers seen at T3. These nine teenagers presented with absence of clinical symptoms, normal PSQ responses, normal clinical and orthodontic evaluation and cephalometric X-rays considered as normal for age and had an ambulatory study performed with a cardio-respiratory monitor (Ambletta™) with oxymetry and peripheral arterial tonometry-(PAT) measuring apnoea and hypopnoeas and "RERAs Embletta". The "RERA Embletta" was based on nasal flow and thoraco-abdominal efforts, with a decrease in nasal flow by at least 30% and an increase in respiratory efforts shown by thoraco-abdominal curves, terminated by evidence of a sympathetic activation indicated by change in the PAT curve. Mouth breathing was monitored with a thermistor.

Cephalometric X-rays were taken in a neutral head position seated with a cephalostat. Cephalometric assessments were performed according to Ricketts parameters [18]. Lateral and frontal cephalometrics were analysed by the orthodontist–craniofacial specialists. For calculation, correction using the Frankfort plane was done. From the many measurements that can be extracted from cephalometric X-rays, several measurements were pre-selected as 'variables of interest' for this retrospective study. They included the measured angles sella-nasion-supradentale or point A (SNA), sella-nasion-infradentale (or pogonion-point B) (SNB), the difference between the two angles: that is, the angle formed by point A-nasion-point B (ANB), the distance mandibular plane-hyoid bone (MP-H), the length of the mandibular ramus (BL) and the posterior airway space (PAS) – defined as the narrowest point
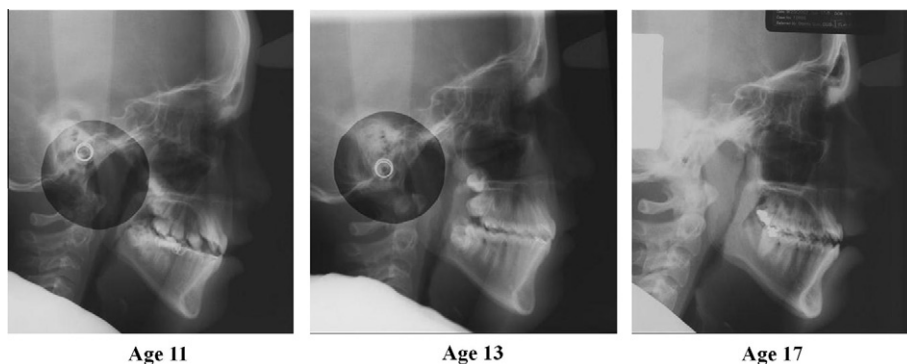
Fig. 3. Cephalometrics changes with age. Another example of the changes in the upper airway anatomy with aging; the teen-ager was treated with nasal CPAP after recurrence of OSA with planning for maxilla-mandibular advancement at late teen-age. Despite presence of symptoms, compliance with nasal CPAP was poor and there was progressive worsening of clinical symptoms and oral-facial presentation.

behind the base of tongue [19]. It was calculated from the anterior point of the velum to the pharyngeal wall (see Figs. 2 and 3).

2.4. Statistical analysis

T-tests for repeated measures were used when comparing two measurements at different ages, and the non-parametric Wilcoxon sign rank test was used for all other comparisons.

3. Results

All children were either Asian or Caucasian. During the survey period of these retrospective evaluations, only 29 children were seen at the three time points and formed the primary study group (see flow chart, Fig. 1). There were nine girls and 20 boys. When seen at the orthodontic clinic at the pubertal time point (T3), nine adolescents had no clinical complaints. Parents reported that they intermittently looked "tired" during the week and three of them had some indication of mild sleep-phase delays with long sleep on weekends; these teenagers had normal responses at PSQ and had normal orthodontic evaluation. The 20 other children had changes on orthodontic imaging and underwent in-laboratory PSG.

The retrospective findings at three different time points from time of diagnosis as a young prepubertal child (T1) till the pubertal evaluation period (T3) for these 29 teenagers are outlined below and in Tables 1–3:

3.1. +Pre-pubertal initial data (T1): (see Table 1)

All 29 children had clinical symptoms and presentation supporting the diagnosis of SDB at the time, and adenotonsillectomy was recommended and performed in all cases following the diagnosis of OSA as confirmed by PSG.

The PSG results before and after adenotonsillectomy are outlined in Table 1.

Following the surgical treatment, clinical evaluation and postoperative PSG, it was felt that these subjects would benefit from RME or bi-maxillary treatments [8]. All children were followed up for about 12 months by orthodontists. They had a PSG performed at the beginning of orthodontic treatment, and again at approximately 1 year (range 10–13 months) after termination of all upper airway treatments (see Table 1). At this time, there were no further recommendations for treatment of SDB in any of the children undergoing orthodontic treatment. However, it was recommended that all children follow-up with their orthodontists to evaluate their bite.

3.2. +Pre-pubertal-orthodontic clinics data (T2)

Orthodontic charts indicate the follow-up of these 29 children between 10 and 11 years of age. At this time point, all 29 children

Table 1

	Initial diagnosis	Post-treatment T and A	Post-treatment orthodontics+	Post treatment final evaluation
Number of subjects	29	29	29	29
Number of boys	20	20	8	20
Age (y) mean ± SD	7.6 ± 1.7	7.8 ± 1.8	7.10 ± 2.0	8.6 ± 2.8
AHI mean ± SD	9 ± 5	3 ± 4	0.5 ± 0.2	0.4 ± 0.4
RDI mean ± SD	15 ± 6.4	7 ± 6	0.8 ± 0.2	0.6 ± 0.5
Lowest SaO2-% mean ± SD	91 ± 2.5	94 ± 3	97 ± 1	98 ± 1.5

Table 2
Recording results in mean age 14 years teen-agers.

Pubertal teen-agers N = 29	No clinical complaints	With clinical complaints
Number of subjects	9	20
Number of boys	2	18
BMI (kg/m^2)	15.9 ± 1.9	15.7 ± 2.1
Age (y) mean ± SD	13.8 ± .9	14.2 ± 1
AHI[**] mean ± SD	0.5 ± 0.2[*]	3.1 ± 1.0
RDI[**] mean ± SD	1.5 ± 1.2[*]	7 ± 1.2
Lowest[**] SaO2-% mean ± SD	97 ± 1%[*]	92.5 ± 1.5

[*] Polygraphic recording with "Embletta-PAT".
[**] Significantly different between group (Wilcoxon test $p = 0.05$).

Table 3
Selected measures from cephalometrics, before puberty and at investigation during puberty.

Cephalometrics	Mean age 11 years	Mean age 14 years
SNA	84.7(1.7)	85.1(2.4)
SNB	79.4(2.65)	79.85(2.65)
H-MP	10.1(3.75)	11.8(9.25)
PAS[*]	9.0(1.85)	6.6(1.85)[*]
BL	76.4 (5.5)	83.2 (5.1)[*]

S = sella.
N = nason.
A, point A: the most posterior midsagittal point on anterior maxillary surface.
B, point B: the most posterior midsagittal point on anterior mandibular surface.
PAS: posterior-airway-space.
H-MP: distance from mandibular plane to hyoid bone.
BL: length of the mandibular ramus.
[*] Wilcoxon test (two tailed) $p = 0.05$.

completed the PSQ and had cephalometric radiographs. All were still prepubertal. PSQ showed no indication of sleep-related problems, parents had no complaint and cephalometric radiographs were reported as normal for age. Selected measures from cephalometric X-rays were calculated at the time of the retrospective analysis by a single specialist and were considered to show normal growth and development (see Table 3).

3.3. +Pubertal (teenagers) data (T3)

When seen at that age, due to the concerns about the past medical history, and also due to the medical-orthodontic environment and collaborative work between the sleep and orthodontic offices, these teenagers were again questioned about any sleep problems, filled out the SDQ and had repeat cephalometric X-rays performed. It was recommended that these patients also consult the associated sleep clinic. As mentioned, these 29 teenagers were the only ones that followed the suggestion.

3.4. Sleep clinic follow-up for 29 teenagers

3.4.1. Clinical symptoms

The sleep clinic follow-up occurred at a mean age of 14.4 ± 0.9 years. Charts indicated that the Tanner stages [20] were stage 2 in one case, stage 3 in 18 cases and stage 4 in 10 cases; none of the teenagers were post-pubertal. The PSQ indicated that teenagers were sleeping in their own bedroom in all cases [21].

Nine teenagers (seven girls and two boys) had no indication of a sleep problem on SDQ, though there was a tendency to go to bed late in all cases without mention of morning or daytime consequences. Parents reported shortened sleep duration during school days and some late to very late awakenings on weekends,

particularly in three of them. There was no clear clinical concern and these teenagers adjusted without problem to sleep schedule changes particularly between school and vacation schedules.

In 20 cases, there were more concerns expressed by parents and in 10 of them parents had specifically raised the issue with their orthodontist. Symptoms were reported on PSQ. Sixteen of these teenagers had difficulties getting up in the morning and 12 showed some degree of symptoms consistent with sleep-phase delay with consequences on weekdays, including napping on the way home from school (eight cases). Interestingly, the complaint of 'sleepiness' was never mentioned, but report of daytime 'fatigue' was mentioned in 11 cases. Poor sleep with nocturnal awakenings was reported in two cases and snoring sufficiently loudly to be noted by family members was present in five cases. Parents expressed their concern about school performance and difficulty to perform in 15 cases. The same 15 teenagers reported difficulty in concentrating.

Clinical examination of upper airway showed absence of tonsils and adenoids in all cases, but six teenagers (two girls, four boys) had deviated septum and five of them had enlargement of inferior nasal inferior turbinates occupying more than 50% of the nasal passage. At examination, all these teenagers (n = 20) had a Friedman 'modified Mallampati' [10] scale score above 2, (while in the nine other teenagers presenting without complaint, sign or symptom of sleep-related problems only one had a score above 2 (=3). Twelve of the 20 teenagers with sleep-related-complaints had the same Friedman scale score of 4, 16 had high and narrow hard palates, 14 had an overjet of more than 2.5 mm and five had an abnormal overbite.

3.4.2. Myofunctional evaluation

Awake myofacial evaluation [22] of the oral–facial region showed that subjects had an abnormally low tongue position in the mouth, 18 were unable to perform appropriately 'clicking' sounds with tongue, 15 were unable to protrude their tongue upwards when asked to try touching their nose with the tip of the tongue; six had difficulties holding a button between their lips and two had difficulties swallowing when drinking fast. At the end of the evaluation by all subjects were scored with abnormal oral–facial muscle tone during wakefulness. All subjects without complaints had been scored as normal at myofunctional testing [22].

3.4.3. Polygraphic and polysomnographic results

Results of nocturnal polygraphy, performed on the nine teenagers without signs or symptoms, and nocturnal polysomnography performed on the last 20 teenagers, are shown in Table 2. This last group of teenagers had a mean time spent in flow limitation of 65 ± 12%, compared to 21 ± 6% in the non-complaining group (See Fig. 4). This measurement was the most abnormal finding at this test performed at the teenage.

3.4.4. Cephalometric-selected-variable evaluation

We evaluated the cephalometric- selected variables at T2 (mean age of 11 years) and no significant difference could be found between the 29 subjects. Results were considered normal for age and growth. There was, however, a change at a mean of 14 years of age. When comparing the measurements obtained on the 20 children with clinical symptoms and SDB by polysomnography at a mean age of 14 years at T2 versus those obtained at T3, a significant decrease in the width of the PAS [19], from 9.15 ± 1.85 to 6.6 ± 1.85 mm was noted (two-tailed $p = 0.05$) (see Table 3 and Figs. 2 and 3). This finding was in contrast with the comparison performed in the nine other children, where no significant change was observed.

The other measurements performed showed no other change (see Table 3). The measurement of the mandibular ramus indicated that all children had growth of the mandible with significant
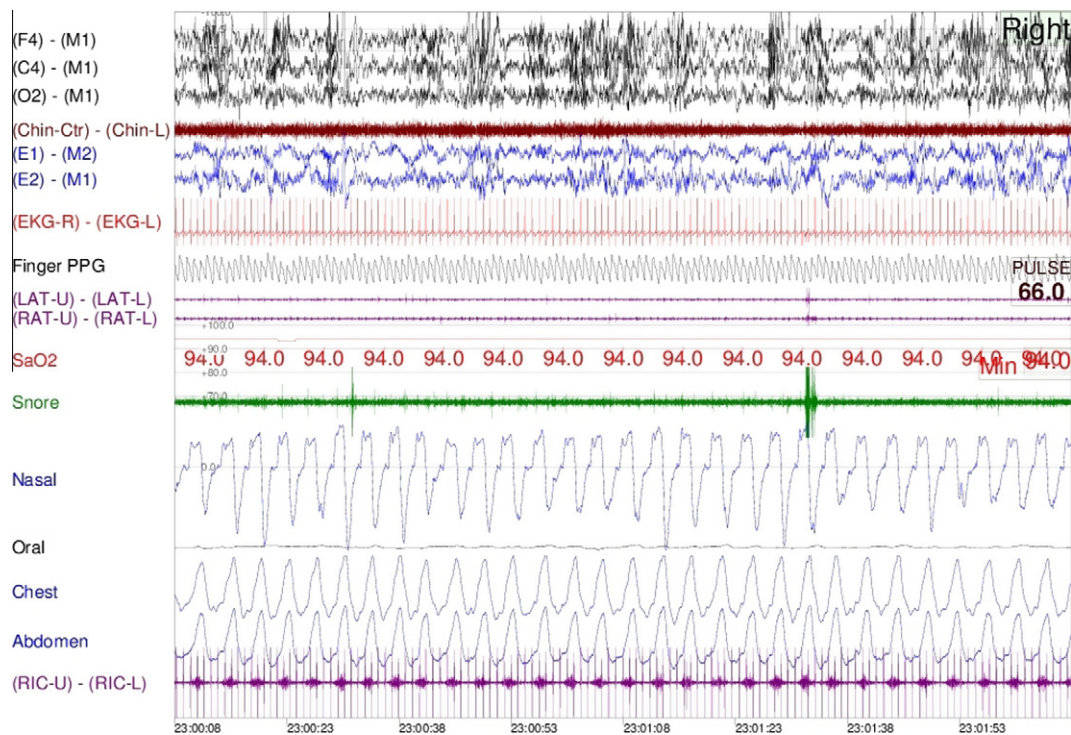
Fig. 4. Flow limitation. Example of 2 min polysomnographic recording with flow limitation in a pubertal teen-ager with clinical symptoms. Note the repetitive high amplitude EEG discharge part of the phaseA of the cyclic alternating system on the EEG leads (1–3 from the top), note the abnormal curve of nasal cannula-pressure-transducer recording on tracing #13 from the top ("nasal"). This tracing is characteristics of ":flow limitation" tracing. Note that oxygen saturation tracing (# 11 from the top) do not change.

change between the mean of 11 years and mean of 14 years of age, and there was no significant difference between the two subgroups of teenagers at a mean of 14 years of age.

In summary, a group of teenagers considered as cured of SDB since their treatment in early childhood and with no clinical complaint and normal clinical oral–facial evaluation had reoccurrence of clinical complaints and presented with abnormal clinical oral–facial and myofunctional examinations [22] and changes in measurement of the PAS [19] at cephalometric X-rays (and to a lesser degree, a lengthening of the MP-H distance [19]. In one case, symptoms and cephalometric changes were documented 1 year apart. The most abnormal polysomnographic findings involved the amount of flow limitation and mouth breathing during sleep. This was associated with an increase in RERAs and to a lesser degree with an increase in AHI. SDB complaints had reoccurred between a mean age of 11 and 14 years.

4. Discussion

This is the first study that illustrates the very long-term evolution of OSA in children after report of adequate treatment of SDB. It suggests that the reappearance of SDB can occur several years following adequate surgical and orthodontic treatment, and that the presence of flow limitation on PSG may be an early indicator of this recurrence in adolescents. Anecdotally, many adult patients with OSA in our clinic report a history of snoring during childhood that resolved or improved after adenotonsillectomy, only to return in their late teens or early twenties. This study provides possible insights that may help to explain this report. In our small study, recurrence of OSA was seen more frequently in boys than in girls and involved a clear decrease in PAS on cephalometrics X-rays (see Figs. 2 and 3). Several studies have shown that children may still present with OSA after adenotonsillectomy, and even after orthodontic treatment, when evaluated with polysomnography [23]. However, the children in our retrospective studies had normal PSGs after performing both treatments. With such results, paediatricians will consider the health problem as resolved, as did sleep specialists at the time.

Our study has all the problems associated with retrospective studies: A large number of children had no follow-up. Unfortunately, this is the reality of clinical practice. This study faced other limitations typically associated with retrospective studies: a small sample size due to inadequate medical records and non-compliant follow-up. In addition, our study gives no indication of the frequency of the recurrence of OSA in individuals considered as "cured" after adenotonsillectomy alone during early childhood, despite the fact that the two prior publications indicate that similar recurrence rates were also noted in pubertal teenagers. The children presented here were followed up by orthodontists, and this may indicate that their anatomy may have been different from other children. A prior systematic evaluation of children using clinical anatomic scales has shown that one mild anatomic oral–facial considered as abnormal was very common in children with OSA, and the involvement of each of these independent anatomic factors in occurrence of OSA was difficult to assess as a single of these abnormal findings was noted in subjects with both normal and abnormal PSG results post-adenotonsillectomy [21], rendering their predictive value very questionable. Only three factors were significantly associated with the need for further treatment including a 3 or 3–4 Friedman-modified-Mallampati score [10], presence of a deviated septum and presence of a small mandible [21]. The children presented here were recommended to have orthodontics post-adenotonsillectomy based on their thorough evaluation and follow-up PSG, indicating the importance of such follow-up test not necessarily performed at many other laboratories. However, once these two treatments had been performed, our children

had normal PSGs. With such results, paediatricians will consider the health problem as resolved, as did sleep specialists at the time.

Consistency of PSG and cephalometric measurement analyses over the years was difficult to determine, though the clinics involved in the study had been in collaboration for years.

In addition, despite the fact that the investigation was performed in clinics accustomed to collaborating, one cannot affirm that recordings were analysed similarly, despite the fact that all evaluations and scoring followed international criteria.

One may have wanted several scorers, particularly for cephalometric analyses, with comparison of obtained results between two or more specialists. The protocol called upon only one blinded reader, based on the belief that any bias presented when performing analyses would be systematic and reflected in all results.

In the teenagers with no complaints and symptoms, normal evaluation and normal cephalometry, a simple home study was judged as clinically justified. However, once again one may argue that tests were not exactly similar (ambulatory vs. laboratory study), a problem linked to the retrospective aspect of the study.

Children evaluated at orthodontic and sleep clinics covered by national health insurance were more likely to have follow-up with referral recommendations as compared to those evaluated in locations where such coverage, particularly orthodontics, was handled by private insurances with separate medical and dental insurance plans.

Finally, no child with adenotonsillectomy alone presented data at the three pre-established time points; follow-up and data in these children were very meagre in the consulted databases as shown in Fig. 1.

Despite the above limitations, this is the first study that addresses the long-term evolution of children with early-in-life OSA and recurrence of SDB. We found documentation of normal PAS [20] at a mean of 11 years with absence of complaints, and presence of complaints and small PAS at a mean age of 14 years. Our study also indicates the presence of flow limitation (see Fig. 4) as an important PSG marker of early recognition of reoccurring SDB; flow limitation was associated with mouth breathing and these two polygraphic patterns were observed in large amounts and showed the largest difference compared to the PSG patterns recorded in the nine other non-complaining subjects with normal PAS at cephalometrics.

Our study indicates that the complaint recurrence was not immediate, it occurred during the pubertal period and after a mean age (11 years) where the oral–facial development has usually reached about 90% of its adult final growth. Our study also shows an important amount of flow limitation in association with recurrence of complaints, previously shown to be associated with abnormal sleep and clinical complaints, and large amount of mouth breathing [24,25]. Orthodontists have long associated mouth breathing with oral–facial hypotonia [22,26]. This was affirmed by the abnormal myofunctional evaluations in our SDB teenagers [22]. Muscle hypotonicity is considered to be somewhat responsible for maxillary and mandibular deficiencies noted in many OSA children [27].

5. Conclusion

Resolution of OSA by PSG and disappearance of clinical symptoms during the prepubertal years after adenotonsillectomy with or without [28] rapid maxillary or bi-mandibular expansion does not necessarily indicate complete resolution of the factors responsible for SDB at later age. Recurrence of clinical complaints and SDB is seen with polysomnographic finding of flow limitation in teenagers. Considering knowledge accumulated since the 1970s on risk

of abnormal maxilla and mandibular growth with abnormal breathing, regular follow-up of children with positive history of SDB should be performed particularly during oral–facial growth.

Conflict of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: http://dx.doi.org/10.1016/j.sleep.2012.08.010.

Acknowledgement

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