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ORIGINAL RESEARCH

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Assessment of the risk of sleep-related breathing disorders in young orthodontic population (6–14 years old) in Southern Italy

Ludovica Nucci[1], Salvatore Cocuzza[2], Ilenia Anastasi[3], Ignazio La Mantia[2], Antonino Maniaci[4], Claudia Malara[2], Antonino Lo Giudice[2,*]

[1]Multidisciplinary Department of Medical-Surgical and Dental Specialties, University of Campania Luigi Vanvitelli, 80138 Napoli, Italy
[2]Section of ORL, Department of Medical, Surgical and Technological Sciences, University Medical Center "G. Rodolico-San Marco", University of Catania, 95123 Catania, Italy
[3]Department of Medical-Surgical Specialties—Section of Pediatric Dentistry, School of Dentistry, University Medical Center "G. Rodolico-San Marco", University of Catania, 95123 Catania, Italy
[4]Section of ORL, University of Enna "Kore", 94100 Enna, Italy

***Correspondence**
antonino.logiudice@unict.it
(Antonino Lo Giudice)

Abstract

Background: Sleep-Related Breathing Disorders is characterized by prolonged upper airway obstruction during sleep. The present study aimed to assess the frequency of Sleep-Related Breathing Disorders in the pediatric orthodontic cohort representative of the Southern Italy population, and to evaluate the prevalence of the type of malocclusion in subjects resulting at risk of Sleep-Related Breathing Disorders. **Methods**: The study sample comprised 364 children, aged 6–14 years who referred to the Department of Orthodontics and Pediatric Dentistry of the University of Catania for orthodontic treatment. Parents were requested to self-administer the Pediatric Sleep Questionnaire questionnaire (PSQ) through Quick Response (QR) code using Google Forms™ before the orthodontic consultation. Subjects who were at high risk of Sleep-Related Breathing Disorders were retrospectively screened for skeletal malocclusion and craniofacial morphologic characteristics. Data were recorded along with cephalometric parameters describing sagittal and vertical skeletal growth pattern and were statistically analyzed using chi-square tests. **Results**: Within the overall sample, 9.89% of the children were at high risk for Sleep-Related Breathing Disorders. Younger children (6–9 years old) were more prone than older children (11–14 years old) to the risk of developing Sleep-Related Breathing Disorders (13.74% *vs.* 7.76%). Boys were significantly at a higher risk for Sleep-Related Breathing Disorders than girls, in particular at younger age. There were no questions that could differentiate a child considered to be at high risk of Sleep-Related Breathing Disorders. **Conclusions**: The present findings would suggest the importance of conducting screenings for Sleep-Related Breathing Disorders in young orthodontic patients and to address individuals identified as high-risk to the sleep-related breathing disorders (SRBD) specialists for additional assessment and treatment.

Keywords

Obstructive sleep apnoea syndrome; Paediatric sleep questionnaire; Sleep breathing disorder; OSA; OSAS

1. Introduction

Sleep-related breathing disorders (SRBD) in children include several conditions that can affect their breathing patterns during sleep, leading to disrupted sleep and potential long-term health issues. SRBD are characterized by instability or complete obstruction of the upper airway during sleep, resulting in impaired ventilation [1]. These disturbances alter the sleep architecture, leading to fragmented sleep, daytime drowsiness, and increasing the risk of morbidities such as hypertension and cardiovascular issues [2]. Obstructive sleep apnea (OSA) is the most common SRBD condition and is characterized by chronic episodes of partial or complete upper airway obstruction [3]. In children, OSA can negatively impact cognitive and behavioral development, affecting their readiness for school [4]. Children with OSA may show deficits in behavior, emotional regulation, scholastic attitude and attention [5]. Also, airway obstruction in pediatric OSA can cause mouth breathing, which may influence head posture and disrupt normal craniofacial and dentoalveolar development. In this regard, OSA has been associated with a significantly high risk of altered craniofacial morphology [6] and malocclusion [7] in pediatric population referred to otorhinolaryngology specialists.

Early diagnosis of SRBD in growing individuals is crucial for preventing or reducing symptoms [8–12]. In this regard, a recent white paper emphasized the importance of diagnosing OSA in children and recommended incorporating OSA screening into orthodontic examinations [13–15]. As for adults, the gold standard for sleep disorder diagnosis is supervised in-laboratory polysomnography (PSG) [16]. While

SRBD and OSA are well-documented in adults, determining the prevalence of SRBD in children is challenging [17]. In-laboratory PSG is impractical for screening due to cost, and several sleep questionnaires have been developed and validated for screening SRBD in children [18, 19]. Among them, the Pediatric Sleep Questionnaire (PSQ) was the first validated tool for assessing SRBD in children with good accuracy for screening and identifying the risk factors for SRBD [20, 21]. For the above-mentioned considerations, orthodontists and pediatric dentists can serve as the frontline in Sleep-Related Breathing Disorders diagnosis since they routinely examine children and adolescents in the daily practice [14].

However, the assessment of the likelihood of developing SRBD through questionnaire-based methodology is contingent upon the educational and socioeconomic background of the parents. Consequently, it becomes imperative to scrutinize the SRBD risk within diverse orthodontic populations. In this regard, the present study aimed to assess the frequency of Sleep-Related Breathing Disorders in the pediatric population referring to the Department of Orthodontics and Pediatric Dentistry of the University of Catania representative of the Southern Italy population, and to evaluate the prevalence of malocclusion in those subjects resulting at risk of OSA. This information would help to enhance awareness of the diagnosis and treatment of obstructive SRBD in children, thereby preventing potential developmental, educational, mental, and oro-functional problems in undiagnosed children. The null hypothesis of the study was the absence of significant differences in the prevalence of high-risk for SRBD within the orthodontic population according to age, gender and growth pattern characteristics.

2. Materials and methods

The study sample comprised 364 growing individuals aged 7–14 years seeking orthodontic consultation at the Department of Orthodontics and Pediatric Dentistry of the University of Catania Italy, during the period between October 2021 and October 2023. We included individuals within 6–14 years of age since this is the general age range of orthodontic population. Also, late adolescents and adults were excluded because the aim of the researchers was to address the analysis on growing individuals. The group included 196 boys with a mean age of 11.75 years and 168 girls with a mean age of 12.75 years. Subjects were also divided into two groups according to two different sub-age ranges, *i.e.*, 6–9 years (Group A) and 11–14 years (Group B). The age distinction between the two groups was established based on evidence indicating that adenoid tissue reaches its peak growth around the age of seven while tonsil size is likely to have diminished by the age of 11 [22, 23]. The study was conducted following Helsinki Declaration on medical protocols and ethics and parental informed consent was obtained via individual signature. The study utilized the Pediatric Sleep Questionnaire (PSQ) developed by Chervin *et al*. [21] at the University of Michigan in its Italian version translated by Ranieri *et al*. [24]. The PSQ aims to identify children at elevated risk of obstructive sleep-related breathing disorders (Sleep-Related Breathing Disorders) and associated symptoms, encompassing phenomena such as snoring, daytime sleepiness and associated behavioral disturbances.

The questionnaire is directed towards the parents of the subjects and consists of 22-item questions divided into three domains: snoring (9 items), sleepiness (7 items) and behavioral (6 items). PSQ investigates symptoms including snoring characteristics such as duration, intensity, frequency, episodes of apnea, mouth breathing, presence of enuresis, excessive sleepiness during the day, headache, weight and growth, symptoms of hyperactivity–impulsivity and inattention. The questionnaire presents a straightforward and concise format, with a response structure of "yes", "no" or "don't know". Affirmative responses are assigned a score of "1", while negative responses are scored as "0". "Don't know" responses are subtracted from the total score. An average score of 0.33 (indicating 33% "yes" responses) or higher indicates an elevated risk for SRBD. Fig. 1 shows a detailed description of the PSQ questionnaire administered for the present study. Each parent was asked to self-administer the questionnaire through QR code using Google Forms™ before the orthodontic consultation. All data related to the PSQ were automatically recorded using a dedicated spreadsheet generated form the Google Form platform.

In addition to the questionnaire, subjects who were at high risk of Sleep-Related Breathing Disorders were retrospectively screened for skeletal malocclusion and craniofacial morphologic characteristics. The following two cephalometric parameters from the analysis of the European Board of Orthodontics were considered for the diagnosis of growth pattern: A-N-Pog° (sagittal diagnosis) and SN-GoGn° (vertical diagnosis). In particular, a dedicated datasheet was created with the diagnostic cephalometric parameters of each patients. The clinical examination and cephalometric analysis was performed by an orthodontist with 15 years of clinical experience (ALG). Finally, the data related to the PSQ and to the orthodontic diagnosis were integrated into a single final datasheet.

A preliminary evaluation of sample size power was performed on 100 subjects (50 in Group A and 50 in Group B), the analysis suggested that 111 patients for each group were required to reach the 80% power to detect a mean difference of 5.1% in the risk of occurrence of SRBD in between both groups, with a confidence level of 95% and a beta error level of 20%.

Preliminary data analysis was performed using the Shapiro-Wilk and Levene's tests to assess data distribution and equality of variance. Since data showed normal data distribution, parametric tests were used. For comparative analysis of prevalence, *i.e.*, distribution of high-risk and low-risk subjects according to age, gender and age sub-groups, the chi-square test was applied. The same tests were used to analyze the distribution of subjects with high-risk of sleep disorder according to skeletal growth pattern. Statistical significance was set at $p < 0.05$. Datasets were analyzed using SPSS® version 24 Statistics software (IBM Corporation, Armonk, NY, USA).

3. Results

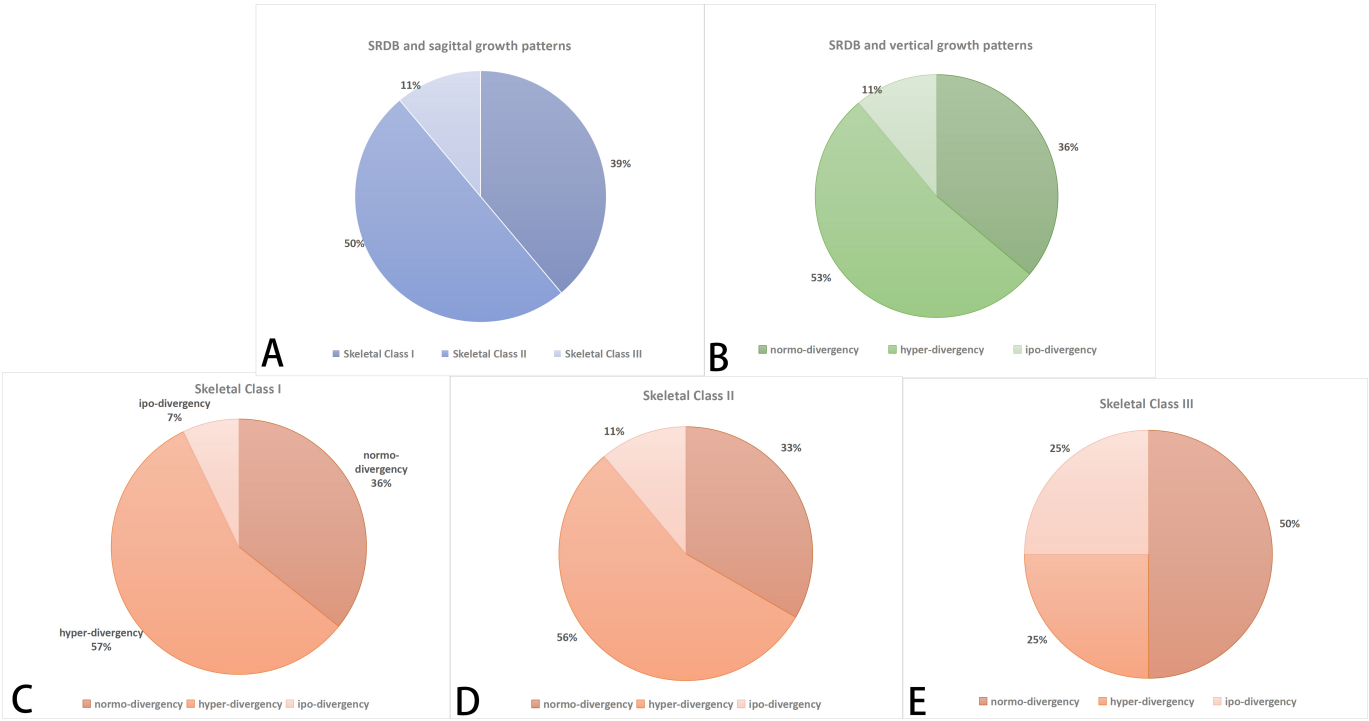

FIGURE 1. Distribution of high-risk subjects for sleep-related breathing disorders according to skeletal malocclusion and facial growing pattern. (A) Sleep-Related Breathing Disorders and sagittal grow pattern; (B) Sleep-Related Breathing Disorders and vertical grow pattern; (C) Sleep-Related Breathing Disorders and sagittal (class I)/vertical grow pattern; (D) Sleep-Related Breathing Disorders and sagittal (class II)/vertical grow pattern; (E) Sleep-Related Breathing Disorders and sagittal (class III)/vertical grow pattern. SRDB: sleep-related breathing disorders.

3.1 Sleep-related breathing disorders and gender

According to the data extracted from PSQ, 36 out of the 364 children (9.89%) were at high risk for Sleep-Related Breathing Disorders (Table 1). Boys were significantly more prone than girls both considering the overall study sample (13.27% *vs.* 5.95%; $p < 0.05$) and isolating the sample at high risk for Sleep-Related Breathing Disorders (72.22% *vs.* 27.77%; $p < 0.05$). In the population with high-risk, males presented younger age compared with females (11.3 *vs.* 13.5 years; $p < 0.05$), instead there was no significant difference in the mean age of males *vs.* females in the total sample (12.2 *vs.* 11.8 years, data not shown).

3.2 Sleep-related breathing disorders according to age groups

Boys and girls were separated into 2 age groups to analyze the relationship between the frequency of Sleep-Related Breathing Disorders risk and age (Table 2). One hundred thirty-two children aged between 6–9 years (Group A) and 232 children aged between 11–14 years (Group B). Younger children (6–9 years old) were more prone than older children (11–14 years old) to the risk of SRBD (13.64% *vs.* 7.76%; $p < 0.05$). There were significantly more boys at high risk in the younger age group compared with girls (19.44% *vs.* 6.67%; $p < 0.05$). However, such differences became smaller when both sexes were compared in the older age group (9.68% *vs.* 5.56; $p < 0.05$). Furthermore, there were no significant differences in the frequency of Sleep-Related Breathing Disorders risk between the girls in the younger *vs.* older age group ($p > 0.05$); instead, the risk for SRBD was significantly different between boys in the younger *vs.* older age group ($p < 0.05$).

3.3 Sleep-related breathing disorders and early malocclusion

According to the skeletal cephalometric analysis, there were no statistically significant differences in the distribution of high-risk individuals among different sagittal growth patterns (Class I, II and III; $p > 0.05$) (Fig. 1A); instead, significant differences were recorded in the distribution of high-risk individuals among different vertical growth patterns, with higher incidence for those subjects featuring hyper-divergency of the mandibular plane ($p < 0.05$) (Fig. 1B). Integrating data from sagittal and vertical classifications, significant differences were recorded only within Class II individuals, with higher incidence for those subjects featuring hyper-divergency of the mandibular plane ($p < 0.05$) (Fig. 1C–E).

3.4 PSQ analytic evaluation

The responses to each question were analyzed to evaluate the (1) number of positive responses, (2) gender and age differences in positive responses between boys and girls, (3) differences in positive responses between age Group A and B.

No significant differences were observed in the rate of positive responses to each question ($p > 0.05$), indicating that no single question effectively identified children at high risk for Sleep-Related Breathing Disorders. In general, questions n. 19, 22, 8, 13, 7 reported more positive answers compared to the

TABLE 1. Data distribution of SRBD risk according to baseline factors.

Factors	Low Risk for SRBD		High Risk for SRBD		Significance
	Mean	SD	Mean	SD	
Age (y)					
Boys	12.2	2.11	11.3*	1.78	$p = 0.019$
Girls	12.4	2.27	13.5*	1.95	
	n	%	n	%	
Gender, n (%)					
Boys	170	86.73	26	13.27**	$p = 0.027$**
Girls	158	94.05	10	5.95**	
Total	328	90.10	36	9.89	

y: years; SD: standard deviation; n: number; SRBD: sleep-related breathing disorders.
**p value based on t-test and set at 0.05; **p value based on chi-square test and set at 0.05.*

TABLE 2. Data distribution of SRBD risk according to the age groups subdivision.

Factors	Low Risk for SRBD		High Risk for SRBD		Total (n)	Significance
	n	%	n	%		
Age Groups						
Group A	114	86.36	18	13.64*	132	$p = 0.009$*
Group B	214	92.24	18	7.76*	232	
Total	328	90.10	36	9.89	364	-
Group A						
Boys	58	80.56	14	19.44*	72	$p = 0.011$*
Girls	56	93.33	4	6.67*	60	
Total	114	86.36	18	13.64	132	
Group B						
Boys	112	90.32	12	9.68*	124	$p = 0.023$*
Girls	102	94.44	6	5.56*	108	
Total	214	92.24	18	7.76	232	

n: number; SD: standard deviation; Group A: subjects aged between 6–9 years; Group B: subjects aged between 11–14 years; SRBD: sleep-related breathing disorders.
**p value based on chi-square test and set at 0.05.*

rest of PSQ's list. As shown in Fig. 2, differences in the amount of positive responses between males and females were found in questions 4, 6, 12, 17, 21. No significant differences were found between the mean age of girls and boys with positive answers for each investigated question ($p > 0.05$). Concerning age range, differences in the amount of positive responses between Group A and B were found in questions 3, 7, 11 and 21 (Fig. 3).

4. Discussion

Screening questionnaires serve as the initial method to explore the potential occurrence of a pathological condition in the overall population, thereby minimizing the societal costs associated with the utilization of diagnostic instruments [25]. Questionnaires may be employed in situations where instrumental diagnosis is impractical, leading to an elevated risk of undetected pathology. In this context, despite the crucial need for the timely identification of SRBD and OSA conditions, the persistent issue of underdiagnosis is mostly related to the requirement for polysomnography carried out in sleep laboratories [16, 26, 27].

The Pediatric Sleep Qestionnaire (PSQ) developed by Chervin *et al.* [21] is a validated tool designed to assess the likelihood of childhood SRBD and associated symptoms. Different educational and socioeconomic background represents the main variable that could influence the screening process for the risk of developing Sleep-Related Breathing Disorders via questionnaires [28]. For this reason, it has been advocated to conduct studies in different pediatric orthodontic populations to comparatively assess the incidence of subjects at risk of SRBD and the indicative questions that may help discriminate individuals at high risk [28]. The present investigation was set on the cohort of orthodontic patients retrieved at the Department of Orthodontics of the University of Catania, with the assumption that it reflects

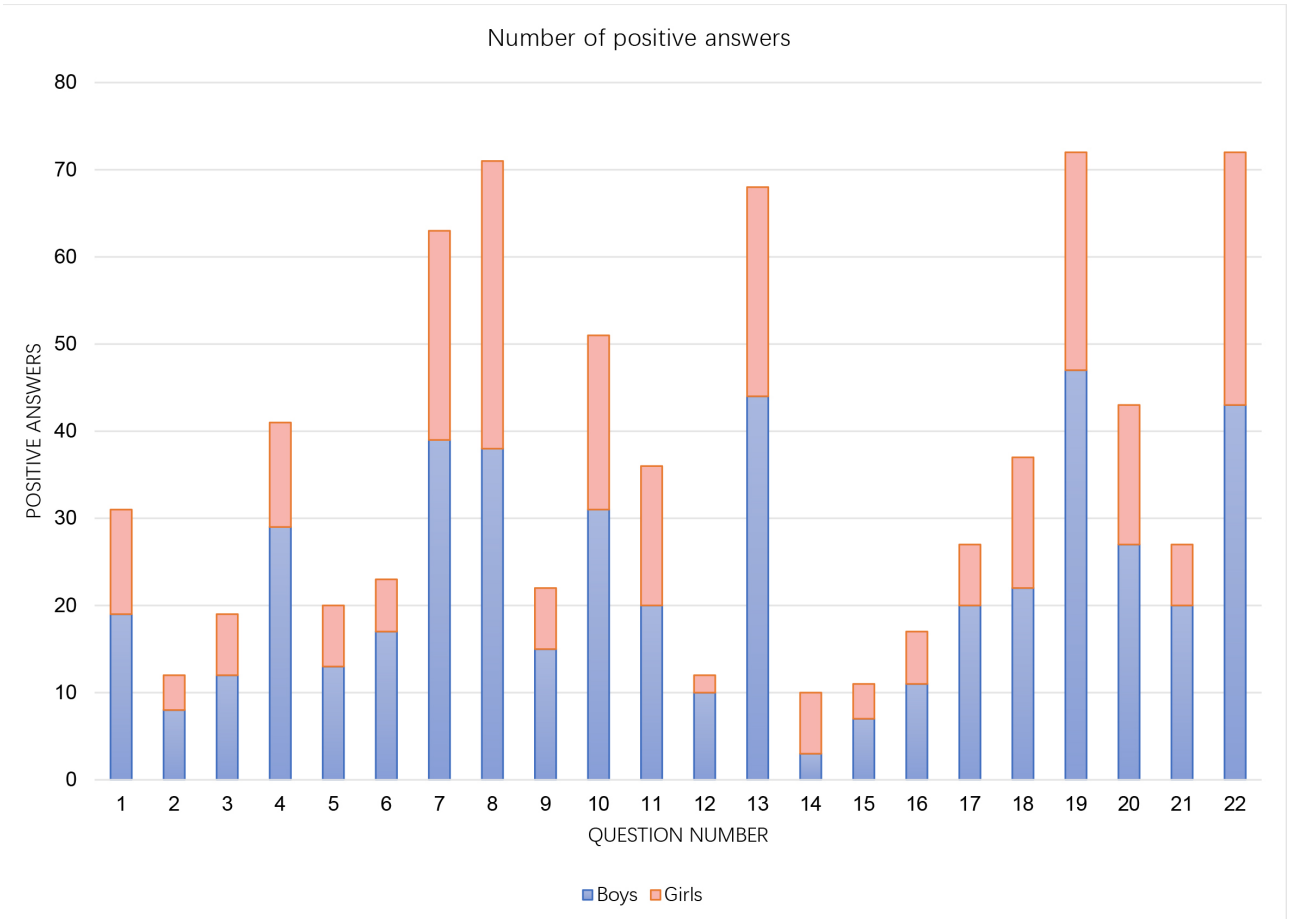

FIGURE 2. Distribution graph of positive answers for each PSQ question, according to gender.

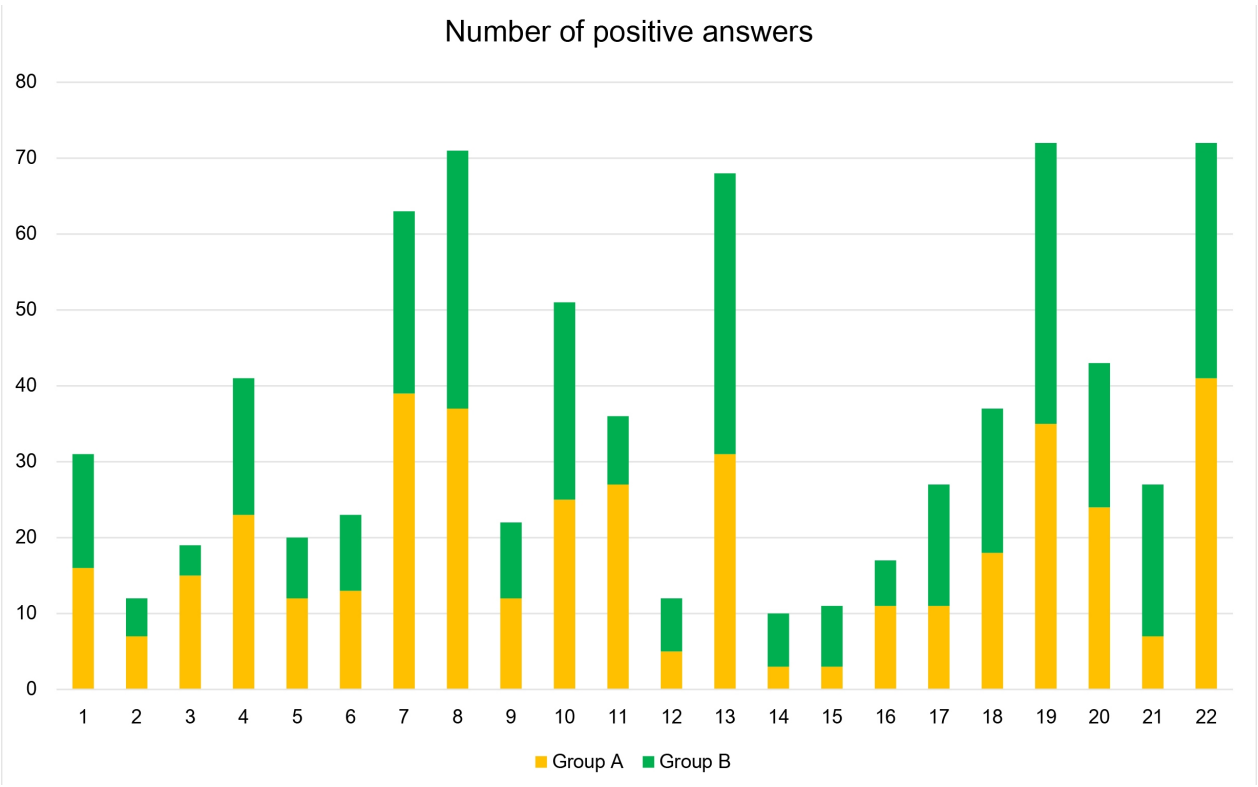

FIGURE 3. Distribution graph of positive answers for each PSQ question, according to age groups.

the young orthodontic population in Southern Italy and the Mediterranean area. To the best of our knowledge, this is the first study in literature that addresses this topic concerning this geographic location.

The findings of the present study revealed that 9.89% of the study sample was at high risk of developing SRBD. This value is within the range obtained extrapolating data from previous evidence of orthodontic populations, respectively of 7.3 % (USA, Ohio) [29] and 10% (Jerusalem, Israel) [28]. Furthermore, the present study revealed that male individuals were at higher risk of developing SRBD compared to females and that the high-risk condition was presented in boys younger than in girls (Male = 11.9 years old, Female = 13.5 years old). Our findings are in agreement with the study performed in the Israelian orthodontic population [28] considering both the different incidences found between gender and the mean age of males and females at high risk; on the contrary, the study performed in Ohio (USA) [27] did not find either gender or mean disparity. Despite the absence of clear consistency among these studies, the present findings and those from the Israelian population concur with previous research indicating higher rates of OSA in males compared to females [30, 31].

Another arguing finding of the present study was that younger individuals (Group A = 6–9 years old) were more prone than older individuals (Group B = 11–14 years old) to be screened as at risk of developing SRBD (13.74% *vs.* 7.76%). This finding may be explained considering previous evidence suggesting that spontaneous improvement of SRBD in children might occur during growth [22, 23]. Specifically, adenoids and the structures within Waldeyer's ring generally grow until the age of 5–7 years, after which they gradually decrease in size [32, 33]. This period also corresponds to the peak incidence of OSA in children [34, 35] and the higher prevalence of adenotonsillar hypertrophy among primary school-age children [34, 36, 37]. As a consequence, it is possible that subjects in Groups A and B could have been exposed to different stages of maturation of the upper airways that could determine a worse respiratory performance in younger individuals (6–9 years old).

Concerning gender differences, there were significantly more boys at high risk in both age groups. Such difference halved in Group B, due to the significant reduction of the incidence of positive scores among male subjects. Consequently, it may be assumed that the difference in upper airway conditions above mentioned could have interested mainly the male gender, as they generally present a delayed growth process compared to females [38]. Also, boys were generally more prone than girls in providing a positive answer to each question of the PSQ questionnaire, and this difference was remarkable in those questions referring to their daily behavior (questions 12, 17 = attention capability, question 21 = hyperactivity) and quality of sleep (question 4 = snoring, question 6 = apnea). Concerning age range, subjects in Group A were generally more prone than subjects in Group B in providing positive response to questions n. 3, 7, 11 which referred to typical nocturnal and daily symptoms of SRBD in children. This finding well align with the above-mentioned from the present study, *i.e.*, the higher possibility for subjects in Group A to be screened as at risk of developing SRBD.

We searched for pertinent questions capable of distinguishing between low-risk and high-risk conditions. In general, questions n. 19, 22, 8, 13 and 7 reported more positive answers compared to the rest of PSQ's list. These questions referred to the hyperactivity behavior of individuals (questions n. 19 and 22), to the nocturnal and daily oral breathing pattern (questions n. 8 and 7), and to the sleepiness at the awakening (question n. 13) and may orientate clinicians assuming a potential risk for SRBD [39, 40]. Nevertheless, since statistical analysis did not identify any specific question that could effectively differentiate a child considered to be at high risk, our findings would confirm that it is necessary to use the entire score of the PSQ to determine if the patient is at high risk [26].

A recent study using PSQ has remarked on the positive answer related to hyperactivity behavior [26], encouraging special attention to the behavioral questions that may help to discriminate if a child is at high risk for SRBD. In the present study, it is possible that parents may not discriminate dysfunctional hyperactivity from similar behavior derived from uncontrolled vivacity, due to different educational backgrounds. As a consequence, in Southern Italy, the behavioral questions of PSQ may not be considered indicative parameters to discriminate subjects at risk of SRD since parents may underestimate the neurocognitive changes that could impair behavior, emotional regulation, school performance, memory and alertness [20, 41].

We collected diagnostic data from cephalometric records of high-risk individuals to identify the craniofacial morphological characteristics potentially associated with the risk of SRBD in growing individuals. Our findings revealed that the incidence of high-risk individuals was higher in the presence of a tendency toward hyper divergency mandibular growing pattern, also in association with class II skeletal diagnosis. These data agree with those reported in a recent well-conducted meta-analysis [6] delineating the craniofacial morphology consistent with pediatric obstructive sleep apnea syndrome (OSAS) and oral breathing pattern, *i.e.*, mandibular retrusion, maxilla-mandibular divergency, increased vertical growth [42], Class II malocclusion [43] and a skeletal maxillary constriction [44–46]. From the clinical perspective, if craniofacial morphology is combined with a medical history of snoring, difficulty in nasal breathing, notable allergies, asthma or obesity, the pediatric dentistry/orthodontist should direct the individual to an otolaryngologist for evaluation. According to the present findings and considering the potential threat that SRBD poses to the well-being and growth of young individuals, it would be of clinical relevance to routinely administer questionnaires in pediatric dentistry/orthodontic clinics to screen high-risk children [43, 47–49], with the final aim to intercept those patients who require additional assessment, diagnostic insights and treatment [50].

5. Limitations

The main limitation of the present study is the analysis of the presumed risk for SRBD based on questionnaire. In this regard, only instrumental examinations can confirm the diagnosis of OSA, thus, providing information about the efficacy of PSQ in predicting SRBD in the pediatric population of

Southern Italy. Also, the study was design as observational-cross sectional survey that does not provide cause-and-effect relationships between variables. Overnight polysomnography is considered the gold standard for diagnosing SRBD and OSA in children who habitually snore, but it is costly and not always accessible. Alternatively, nocturnal oximetry has proven to be a valuable tool that can assist in treatment decisions when polysomnography is unavailable [22]. In this regard, future studies that integrate a first-line screening process with PSQ and a second-line diagnostic validation using PSG or nocturnal oximetry are encouraged to clarify the predictability of PSQ in discriminating subjects affected by OSA and the association between this pathology and malocclusion/skeletal pattern in Southern Italian population.

6. Conclusions

The PSQ examination showed that 9.89% of the children aged 6–14 years, who were referred for orthodontic consultation were at high risk for SRBD.

Younger age children (6–9 years old) were more prone than older age children (11–14 years old) to the risk of developing SRBD (13.74% *vs.* 7.76%).

Boys were significantly at a higher risk for SRBD than girls and at a younger age.

Individuals featuring hyper-divergency of the mandibular plane were at high risk for SRBD compared to normo-divergent and hypo-divergent individuals.

There were no questions that could differentiate a child considered to be at high risk of SRBD, thus, it is necessary to use the entire score of the PSQ for screening evaluation.

AVAILABILITY OF DATA AND MATERIALS

The data presented in this study are available on reasonable request from the corresponding author.

AUTHOR CONTRIBUTIONS

ALG—designed the research study; wrote the manuscript; performed the research. IA—co-wrote the manuscript and performed statistical analysis. LN—performed statistical analysis. CM, AM, ILM, SC—analyzed the data. All authors read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The study was approved by the Ethical Committee of Catania—AMMOC project 15472022/PO (12 December 2022). The parents signed appropriate consensus form related to consensus to participate to the study.

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CONFLICT OF INTEREST

The authors declare no conflict of interest. Antonino Lo Giudice is serving as one of the Editorial Board members of this journal. We declare that Antonino Lo Giudice had no involvement in the peer review of this article and has no access to information regarding its peer review. Full responsibility for the editorial process for this article was delegated to APG.

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