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ORIGINAL ARTICLE

Lateral facial profile may reveal the risk for sleep disordered breathing in children—The PANIC-study

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Abstract

Objective. To evaluate the lateral view photography of the face as a tool for assessing morphological properties (i.e. facial convexity) as a risk factor for sleep disordered breathing (SDB) in children and to test how reliably oral health and non-oral healthcare professionals can visually discern the lateral profile of the face from the photographs. ***Materials and methods.*** The present study sample consisted of 382 children 6–8 years of age who were participants in the Physical Activity and Nutrition in Children (PANIC) Study. Sleep was assessed by a sleep questionnaire administered by the parents. SDB was defined as apnoeas, frequent or loud snoring or nocturnal mouth breathing observed by the parents. The facial convexity was assessed with three different methods. First, it was clinically evaluated by the reference orthodontist (T.I.). Second, lateral view photographs were taken to visually sub-divide the facial profile into convex, normal or concave. The photos were examined by a reference orthodontist and seven different healthcare professionals who work with children and also by a dental student. The inter- and intra-examiner consistencies were calculated by Kappa statistics. Three soft tissue landmarks of the facial profile, soft tissue Glabella (G̀), Subnasale (Sn) and soft tissue Pogonion (Pg̀) were digitally identified to analyze convexity of the face and the intra-examiner reproducibility of the reference orthodontist was determined by calculating intra-class correlation coefficients (ICCs). The third way to express the convexity of the face was to calculate the angle of facial convexity (G̀-Sn-Pg̀) and to group it into quintiles. For analysis the lowest quintile (≤164.2°) was set to represent the most convex facial profile. ***Results.*** The prevalence of the SDB in children with the most convex profiles expressed with the lowest quintile of the angle G̀-Sn-Pg̀ (≤164.2°) was almost 2-fold (14.5%) compared to those with normal profile (8.1%) ($p = 0.084$). The inter-examiner Kappa values between the reference orthodontist and the other examiners for visually assessing the facial profile with the photographs ranged from poor-to-moderate (0.000–0.579). The best Kappa values were achieved between the two orthodontists (0.579). The intra-examiner Kappa value of the reference orthodontist for assessing the profiles was 0.920, with the agreement of 93.3%. In the ICC and its 95% CI between the two digital measurements, the angles of convexity of the facial profile (G̀-Sn-Pg̀) of the reference orthodontist were 0.980 and 0.951–0.992. ***Conclusion.*** In addition to orthodontists, it would be advantageous if also other healthcare professionals could play a key role in identifying certain risk features for SDB. However, the present results indicate that, in order to recognize the morphological risk for SDB, one would need to be trained for the purpose and, as well, needs sufficient knowledge of the growth and development of the face.

Key Words: *SDB, convex profile, children, lateral facial photographs*

Introduction

Sleep disordered breathing (SDB) is an alarmingly expanding, chronic and progressive condition and is mostly attributable to the increased incidence of overweight and obesity [1]. SDB is a spectrum of symptoms ranging from habitual snoring to obstructive sleep apnea syndrome (OSAS). Although SDB is

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nowadays recognized more often, it is a strongly under-diagnosed condition. The pathophysiology, diagnostics and treatment of SDB among children is still a matter of debate.

There is a growing body of evidence which associates SDB in children with other health problems, such as day-time hyperactivity, symptoms reminiscent of Attention Deficit Hyperactivity Disorder and other behavioral and learning difficulties. Further, night-time enuresis, systemic low-grade inflammation, metabolic syndrome as well as compromised somatic growth and development are well known consequences of untreated SDB [2].

Our recent study revealed that abnormal craniofacial morphology, especially facial convexity, but surprisingly not excess body adiposity, was associated with an increased risk for SDB in children 6–8 years of age [3]. This finding is supported by the results of several other studies in children [4,5]. The impact of body adiposity on sleep problems may increase with age [6] and in adults obesity is the most important risk factor for SDB.

The traditional evaluation of craniofacial morphology can be demanding. The visual evaluation of facial profile is a routine part of the examination of an orthodontic patient. However, the accuracy of evaluation may depend on the experience of the investigator. This examination is usually completed with a roentgen-cephalometric analysis which provides an exact depiction of craniofacial morphology. Because of radiation-hygienic reasons, it would be advantageous to develop methods not involving x-ray imaging, especially for children.

If one wishes to prevent the development or worsening of SDB and related diseases, it is essential to focus on the early diagnosis and treatment of SDB. This goal can only be reached by the better and earlier recognition of the condition, not only by oral health, but, beneficially, also by other healthcare professionals who work with children. There is an obvious need to devise an epidemiological, low-cost and non-invasive method for screening the typical features of a child with SDB. Previous studies have focused on the associations of orofacial skeletal and soft tissue structures with SDB [4,5,7]. Our recent study [3] investigated the craniofacial morphology, assessed at a clinical examination, as a risk factor for SDB for epidemiological screening purposes.

The aim of the present study was to evaluate the lateral view photograph of the face as a tool for assessing morphological properties (i.e. facial convexity) as a risk factor for sleep disordered breathing (SDB) in children and to investigate how reliably a group of healthcare professionals can visually observe the profile of children from photographs to assess the convexity of their faces. We also studied the feasibility of the use of photography for the visual observation of facial profile by testing the intra- and inter-examiner consistency of the method.

Materials and methods

The present analyses are based on the baseline data of the Physical Activity and Nutrition in Children (PANIC) Study, which is a controlled exercise and diet intervention study in a representative population sample of 512 girls and boys aged 6–8 years examined at baseline in 2007–2009. Complete data on variables needed in the analyses were available for 382 (75%) of all 512 children [8]. The mean (median, SD) age of the children was 7.6 (7.6, 0.4) years. The study protocol was approved by the Research Ethics Committee of the Hospital District of Northern Savo. Both children and their parents provided written informed consent for using the whole study data, including the facial photographs.

Sleep was assessed by a sleep questionnaire. The questions were based on validated Finnish questionnaires that have been used to screen for sleep disturbances and SDB [9]. The parents filled out the questions regarding the child's quantity and quality of sleep, symptoms of SDB and upper airway infections and previous operative treatments, such as adenotonsillectomy. SDB was defined as apnoeas, frequent or loud snoring or nocturnal mouth breathing observed by the parents. Of all 382 children, 33 (8.6%) had SDB and there was no statistically significant difference in the prevalence between the genders. Of the children, 29 (7.6%) snored in certain positions, 26 (6.8%) had nocturnal mouth breathing, 20 (5.3%) snored loudly, eight (2.1%) snored frequently and one (0.3%) had witnessed apnoeas. Clinically, the facial profile was assessed visually, as described in detail previously [3]. One third (30%) of the children's profiles were calssified as convex.

A standardized method was used for photographing the participants. The photos were taken using a Canon EOS 300D digital camera in the lateral projection of the face at a distance of 2 m. The children were standing in a natural head position [10,11] looking out of a window with a little bubble level taped on their temple to control the correct head position.

The paper prints of the photos were gathered into a folder and a short instruction guide along with the profile figures was formulated. The folder was examined first by a reference orthodontist (T.I.) and then by seven other healthcare professionals who work with children, i.e. another orthodontist, a dentist with extensive orthodontic experience, a general dental practitioner, an otorhinolaryngologist, a pediatrician, an oral hygienist and a public health nurse. In addition, a dental student examined the folder. The observers classified visually the profiles as normal/mildly convex, clearly convex or straight/concave based on three model pictures (Figure 1).

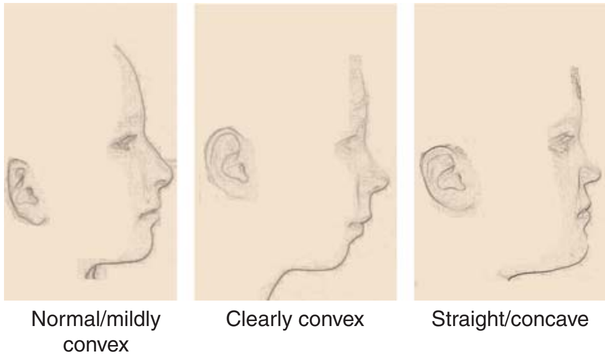

Figure 1. Formula of normal/mildly convex, evidently convex and straight/concave profiles and instructions to the observers.

The photographic data were digitized and analyzed by WinCeph® 8.0 software in order to analyze the profiles more objectively and, further, to compare the subjective and objective assessment. Soft tissue landmarks Glabella (G), Subnasale (Sn) and Pogonion (Pg′) were digitally identified to calculate the angle G-Sn-Pg′, indicating the angle of facial convexity [12] (Figure 2).

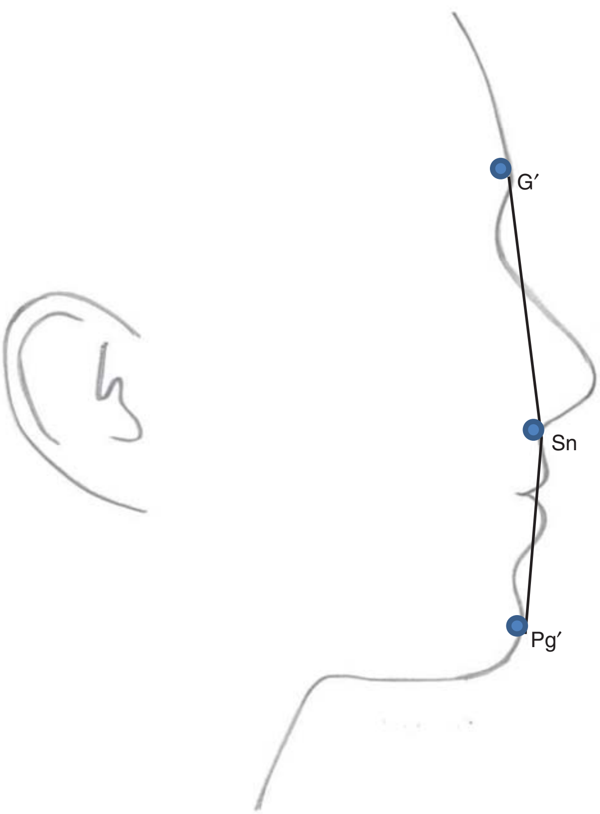

Figure 2. Digitized soft tissue landmarks and lines on lateral facial photographs. (1) Soft tissue glabella (G); (2) Soft tissue glabella reflected on true vertical line (G); (3) Pronasale (Prn); (4) Columnella (Cm); (5) Subnasale (Sn′); (6) Subnasale reflected on true vertical line (Sn′); (7) Labii superior (Ls); (8) Soft tissue pogonion (Pg′); (9) Soft tissue menton (Me′); (10) Soft tissue menton reflected on true vertical line (Me′); (11) Soft tissue porion (Po′); (12) Jaw line; (13) Neck line.

Statistical methods

The angle between soft tissue Glabella, Subnasale and soft tissue Pogonion (G-Sn-Pg′), describing the morphology of facial profile, was grouped into quintiles. For analysis the lowest quintile (≤164.2°) was set to represent the most convex facial profile. Chi-square statistics were used to compare facial convexity between children with and without the SDB.

The inter-examiner reproducibility of the measures of the convexity of the facial profile with the photographs (normal/mildly convex, clearly convex, straight/concave) was studied by comparing the assessments of the other examiners with those of the reference orthodontist using Kappa values and the proportions of agreement [13].

The intra-examiner reproducibility of measures of the angle of convexity of the facial profile (G-Sn-Pg′) was investigated by comparing the first and second assessment 2 weeks apart of the reference orthodontist using intra-class correlation coefficient (ICC) and its 95% confidence interval (CI).

All statistical analyses were performed using the Statistical Package for Social Sciences (SPSS), Version 19.0 (SPSS Inc., Chicago, IL).

Results

The prevalence of the SDB in children with the most convex profiles expressed with the lowest quintile of the angle G-Sn-Pg′ (≤164.2°) was almost 2-fold (14.5%) compared to those with normal profile (8.1%) (p = 0.084). Of all the profiles, which the reference orthodontist clinically classifield as convex, 67.1% were the most convex profiles expressed with the lowest quintile of the angle G-Sn-Pg′. The convex profiles were assessed visually with the photographs by the same examiner, 64.3% were in the lowest quintile.

The Kappa values for the measures of facial profile with the photographs between the reference orthodontist and the other healthcare professionals varied between 0.000–0.579 and the proportion of agreement varied between 55.6–78.8% (Figure 3). The highest Kappa value (0.579) and the highest proportion of agreement (78.8%) were observed between the two orthodontists. The Kappa value for the repeated measurements of the facial profile of the reference orthodontist was 0.920 and the proportion of agreement was 93.3%. The Kappa values for the first 200 photos and the last 200 photos did not differ. The time spent in observing all 382 photographs varied between 25–120 min among the healthcare professionals. The observation time was not associated with the results (data not shown).

The ICC and its 95% CI between the two measurements of the angle of convexity of the facial profile (G-Sn-Pg′) of the reference orthodontist were 0.980 and 0.951–0.992.

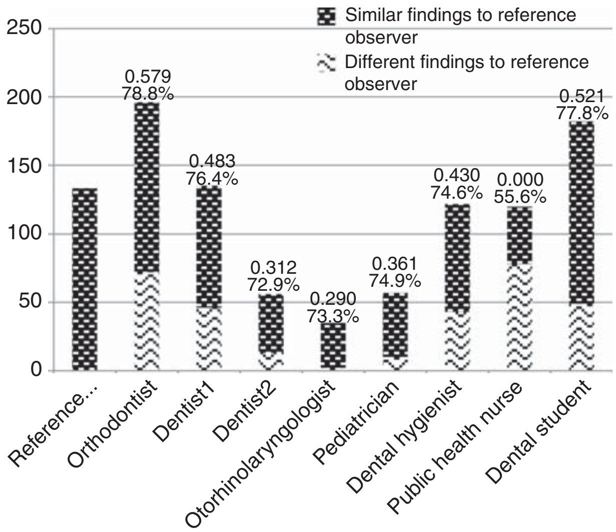

Figure 3. Number of evidently convex profile identified by eight observers from facial photographs (total n = 382). Inter-examiner kappa-values and agreement percentages to reference observer on the top of the bars.

Discussion

The present study highlights the clinical challenges encountered in the diagnostics of craniofacial features related to SDB among children. Our previous study [3] showed that abnormal craniofacial morphology, but not excess body fat, is associated with an increased risk of having SDB in 6–8 year old children. A simple model of necessary clinical examinations (including facial profile) was recommended to recognize children with an increased risk of SDB. In the present study we evaluated the feasibility of lateral facial photographs in the assessment of the convexity of the face. The task was performed by a group of examiners including various healthcare professionals and a dental student and their results were compared to those of an experienced orthodontist. It was apparent that the outcome of the method was subject to extensive variability, i.e. the test as such was not reliable since the Kappa values ranged from poor-to-moderate. However, as expected, the best agreement was found between the assessments of the orthodontists. The inspection of the face is a routine part of a standard orthodontic examination and it strives to estimate the sagittal and vertical characteristics of the face. However, if one wishes to conduct an exact craniofacial examination, the lateral cephalogram is recommended. Even though the visual inspection in the clinical situation is three-dimensional and can be linked with the records from the anamnestic information and clinical examination, it is obvious that an orthodontist can recognize certain facial landmarks, even in the two-dimensional photograph. Nowadays, in most orthodontic cases, pre-treatment and post-treatment photographs are taken as part of treatment planning and for analyzing the outcome of the treatment. In situations when cephalograms are not available, a facial photograph may represent an important diagnostic tool [14].

The angle of convexity of the facial profile was described in 1980 [12] and it was formed by the soft tissue Glabella, Subnasale and soft tissue Pogonion (G-Sn-Pg̀). We used that angle and its lowest quintile as the most objective variable to express the convexity of the face. More than two thirds of the profiles that the reference orthodontist considered visually (both in clinical situation and with the photographs) as convex were also convex by this objective classification. The result indicates that even visual assessment can be valuable for the identification of the convex profiles.

As far as we are aware, no previous studies have evaluated how different healthcare professionals are able to recognize the features of the face. Because in the present study we had only one person representing each health professonal, it was impossible to make straightforward conclusions of the ability of different professionals to evaluate the facial profiles on the basis of the lateral 2-dimensional photographs. However, according to our results, it was difficult for this selected group of health professionals to differentiate clearly convex and normal/concave face profiles except for the orthodontists who have special experience and training for the purpose. The normal growth of the face, both skeletal and soft tissues, occurs in parallel with the somatic growth. However, the growth of the mandible is a slower and longer process than that of the maxilla [15,16]. The facial convexity is an age-related phenomenon and the profile of a prepubertal child usually seems mildly convex [17]. Some of the present observers commented that overweight was the main problem they encountered in the assessment of the facial profile. As found in our previous study in the same study population of children, 15.6% of the girls and 11.4% of the boys were overweight or obese [3]. The presence of excess adipose tissue on the face may obscure the real sagittal facial morphology and among children 6–8 years of age some childish plumpness is still quite common. Furthermore, the craniofacial growth of obese children has been reported to differ from that of lean children [18,19].

In the present study the observers were not calibrated. The introduction to the observation process was mostly based on three different figures of profiles with a short instruction on how to conduct the classification. It can be speculated that, with more comprehensive practice of how to assess the profiles, the results might have been better. In an attempt to see if there was any learning in the process we also compared the Kappa values for the first 200 photos to those for the last 200, but we found no improvement in Kappa values. Further, as mentioned before, a limitation of our study is that there was only one representative of each professional and, therefore, the interpretation of the results must be careful and larger-scale studies on the topic are clearly warranted.

The cephalometric analysis with a lateral roentgenogram is needed for a careful examination of the craniofacial morphology and special cephalometric features of the SDB children are well known [20,21]. However, there are numerous studies showing that the cephalometric variables are relatively strongly correlated with photographic analysis and, with some limitations, the sagittal and vertical relationships of the face can be defined from a lateral facial photo [22,23]. Furthermore, it has been reported that the head position and the craniofacial morphology are associated [24]. Certain anamnestic features such as asthma and allergies are suggested to influence the head position [25] and, thus, are likely to interfere with the the evaluation of the facial profile. This should be taken into consideration in all population-based studies. In the present study, the position of the head was standardized with a bubble level.

Craniofacial morphology has been a routine part of the clinical examination in oral health clinics, especially in orthodontics. General dental practitioners, oral hygienists and dental nurses can consult an orthodontist when they suspect an abnormal craniofacial morphology. Given the current knowledge of the detrimental effects of SDB on overall health, they should be encouraged to refer children at risk of SDB for more detailed anamnestic information and further examinations. In the future, it would be desirable to expand the information about craniofacial risk factors also to other public child health clinics. In particular, it would be useful if public health nurses were able to identify children at the increased risk of SDB. However, comprehensive training of any individual would be needed before he/she could achieve the necessary skills for the identification of children with a risk of SDB on the basis of their facial morphology. In this context, we aim to develop and test a more accurate clinical tool for identifying high risk children. It might be possible that, with training, more healthcare professionals could become proficient, which would improve the early diagnostics of pediatric SDB.

Craniofacial and occlusal development evolves in childhood. Adult patients with SDB seem to display abnormalities in craniofacial morphology and dental occlusion similar to those found in children with sleep-related breathing disorders [4,26]. In children, it would be most advantageous to be able to predict future sleep apnea, the most detrimental manifestation of SDB. This raises the question of whether adulthood SDB could be prevented in some individuals by providing appropriate orthodontic treatment already in childhood [27]. Children with craniofacial risk factors, for example a convex facial profile, could be candidates for early intervention and possible orthodontic treatment to prevent the progression of SDB in later life.

Conclusions

The results indicate that recognizing craniofacial morphology as a risk factor for SDB requires a basic knowledge about how the face grows and develops and also good clinical experience in working with children. Most likely the anamnestic information and lateral facial profile analysis together could predict more accurately OSAS than either one separately do. It would be beneficial if, in addition to oral health professionals, also other healthcare professionals would be trained to identify features related to the increased risk of SDB. Children with a convex facial profile could be candidates for early intervention and orthodontic treatment to prevent the progression of SDB later in life.

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