Craniofacial morphology in preschool children with obstructive sleep apnoea syndrome

ABSTRACT

Aim Obstructive sleep apnoea syndrome (OSAS) is a common problem in children. It is characterised by a combination of partial airway obstruction associated with hypoxemia and hypoventilation and intermittent obstructive apnoea, which disrupts normal ventilation and sleep. The aim of the study was to evaluate the craniofacial features of preschool children with polysomnographic diagnosis of OSAS, using measurements from standardized lateral cephalograms according to the floating norms cephalometric analysis. Materials and methods 21 untreated caucasian children (mean age of 4.57 +/-0.6) with complete deciduous dentition were included in this study. All the subjects had diagnosis of OSAS with a positive RDI. Pretreatment cephalometric radiographs were evaluated. Statistical method Descriptive statistics includes mean and standard deviation of the cephalometric variables. Conclusion The present study showed that OSAS preschool children showed a skeletal Class II pattern with retrognathic mandible and increased skeletal divergency. Key words: Obstructive sleep apnoea syndrome; Preschool children; Cephalometric analysis; Craniofacial morphology.

Introduction

Obstructive sleep apnoea syndrome (OSAS) is a common problem in children. It is characterised by a combination of partial airway obstruction associated with hypoxemia and hypoventilation and intermittent obstructive apnoea, which disrupts normal ventilation and sleep [Chopo et al., 2004; Goldbart and Tal, 2008; Marcus, 2001; Rosen, 1996]. Various craniofacial abnormalities have been reported as predisposing factors such as retrognatism of the maxilla and the mandible, increased lower facial height [Erler and Paditz, 2004; Nordeland et al., 1998], larger craniofacial angulation [Solow et al., 1993] elongated soft palate, decreased posterior airway space and inferior positioned hyoid bone [Lowe et al., 1986; Tangurson et al., 1995; Tangurson et al., 1995].

Diagnosis is based on medical history, physical examination and tests confirming the presence and severity of the upper airway obstruction; however, the gold standard method of assessment is the overnight polysomnography (PSG) [Chan et al., 2004; Jurado Ramos et al., 2006; Rosen, 1996]. Although many parameters are measured with PSG, the respiratory disturbance index (RDI) is the parameter most commonly used to gauge the presence or absence of disease and to assess its severity; the RDI reflects the average number of apnoeas plus hypopnoeas observed per hour of sleep and it is usually derived by the number of respiratory disturbances divided by the number of hours slept [Redline et al., 2000].

Craniofacial anatomy is said to play a role in OSAS and it has been suggested that cephalometry could be an adjunctive procedure for screening anatomical abnormalities in patients with OSAS [Kawashima et al., 2000; Pracharktam et al., 1996; Salles et al., 2005].

Many studies in fact have used cephalometric analysis to assess the craniofacial morphological features of adult and young populations with OSAS, but few of them enrolled children of very young age [Agren et al., 1998; Özdemir et al., 2004; Zettergren-Wijk et al., 2006]. The aim of the present study is to evaluate the craniofacial features of preschool children with complete deciduous dentition and polysomnographic diagnosis of OSAS, using cephalometric measurements performed on standardized lateral cephalograms.

Consent of the parents was obtained prior to the study.

Materials and methods

Subjects

A sample of 21 untreated Caucasian children with complete deciduous dentition (14 males and 7 females), with a mean age of 4.57 +/-0.6 (range 3.11-5.9 years) was selected from the files of the Dental Clinic, University of Rome “La Sapienza”, Sant’Andrea Hospital.

All subjects had a history of disturbed sleep characterised by recurrent apnoeic periods with heavy snoring and had previously been referred to the Paediatric Sleep Disorder Center (University of Rome “La Sapienza”, Sant’Andrea Hospital), where they underwent a standard overnight polysomnographic study, under the surveillance of a trained sleep laboratory technician, and were diagnosed OSAS with a positive RDI.

Cephalometric data

Cephalometric radiographs were obtained before treatment. The cephalometric head plates used in this study were taken with the teeth in occlusion. A cephalostat was used to keep the subject’s head in a
Results

All the subjects have been evaluated by means of polysomnography during a whole night. The RDI was used to quantify the presence of the disease and to assess its severity. A positive RDI (RDI: 7.94 +/- 7.30) confirmed the diagnosis of OSAS.

The mean and the standard deviation of the cephalometric variables of the OSAS children are given in Table 1a, and of the Tollaro’s sample in Table 1b [Tollaro et al., 1996].

According to the Wiggle diagram the cephalometric variables of OSAS preschool children, when compared to a sample of children with ideal occlusion, are in the range of the standard deviation, close to the mean value, with the exception of the SNB, which is close to the left limit and for the ML-NSL which is out of the diagram right limit and thus larger than expected.

As far as the facial type is concerned, the cephalometric values plotted on the floating norms box show the maxilla in the orthognathic range (SNA: 79.63 +/- 2.64), the mandible with a slight retrognathic tendency (SNB: 74.00 +/- 3.08) that determines a distal sagittal basal relationship between the jaws (ANB: 5.55 +/- 1.86) and an orthognathic cranial base angle (NSBa: 132.32 +/- 5.64).

In the vertical plane the angle NL-NSL (NL-NSL: 6.71 +/- 2.04), is harmonious with the degree of maxillary prognathism, whereas the angle ML-NSL (ML-NSL: 39.11 +/- 4.76) is retrognathic and thus disharmonious with the other cephalometric variables with the exception of SNB.

Position, so that the Frankfort horizontal line was parallel to the floor during exposure. Each cephalogram was traced and the following angles SNA, SNB, ANB, NL-NSL, ML-NSL, NSBa, were measured with an accuracy of 0.5° (Fig.1).

Statistical method

Descriptive statistics includes mean and standard deviation of the cephalometric variables.

For all subjects the variables were measured twice by the same observer (AM) and the double measurements were used to assess the measurement error by means of Dahlberg’s formula [Dahlberg, 1940]. The measurements error in degree was 0.56 for SNA, 0.42 for SNB, 0.47 for ANB, 0.87 for NSBa, 0.83 for NL-NSL and 0.80 for ML-NSL.

The variables were compared by means of a Wiggle diagram (Fig. 2), with the cephalometric measurements of a sample of children with ideal occlusion, full deciduous dentition, mean age 5.67 +/- 0.82 [Tollaro et al., 1996], and the floating norms box for the deciduous dentition was used to describe the facial type and to assess the harmony among craniofacial variables (Fig. 3) [Hasund A., 1977; Tollaro et al., 1996].

FIG. 1 - Cephalometric variables: SNA maxillary prognathism, SNB mandibular prognathism, NL-NSL maxillary inclination relative to the anterior cranial base, ML-NSL mandibular inclination relative to the anterior cranial base, NSBa cranial base angle.

FIG. 2 - Wiggle diagram comparing OSAS preschool children cephalometric variables (-----) to Tollaro’s standards for deciduous dentition with normal occlusion (SNA 79.88 +/- 2.91, SNB 76.35 +/- 2.85, NL-NSL 7.94 +/- 2.75, ML-NSL 35.23 +/- 3.27, NSBa 131.56 +/- 4.44) (______) [Tollaro et al., 1996].

FIG. 3 - Cephalometric box of the floating norms in deciduous dentition, for orthognathic, retrognathic and prognathic face types. The template inside the box represents the range of variability for each variable; in the harmonious face the variables values should be located inside the template. The template can be shifted upwards and downwards according to the individual case, if one or more values lie outside the template they will indicate a disharmonious deviation in the growth facial pattern.
Discussion

OSAS is a breathing disorder during sleep characterised by prolonged partial obstruction and/or intermittent complete obstruction of the upper airway that disrupts normal ventilation and pattern during sleep.

There is a continuous interaction between airway patency and maxillomandibular growth [Guilleminault and Stoochs, 1990]. The developing facial skeleton may be influenced by changes in muscle function and extended head posture associated with prolonged airway obstruction and mouth breathing due to adenoidal and tonsillar hypertrophy [Linder Aronson, 1975; Bresolin et al., 1984]. In fact children with OSAS seem to have narrower maxillary width and longer dental arches than non-affected children [Löfstrand-Tideström et al., 1999]. Guilleminault et al. found that a small chin, a steep mandibular plane, a retroposition of the mandible, a long face, a high hard palate, and an elongated soft palate were common among children with obstructed breathing [Guilleminault et al., 1996].

OSA children also have abnormal nocturnal growth hormone secretion (GH) which may contribute to disturbed craniofacial growth [Peltomäki et al., 2007].

Cephalometric analysis is useful as a screening test to characterise skeletal and soft tissue relationships in children having OSAS. In literature many studies have been done to assess the craniofacial architecture of subjects with OSAS but few of them enrolled young children with deciduous dentition [Battagel and LEstrange, 1996; Battagel et al., 2000; Cozza et al., 2004; Kulnis et al., 2000; Pae and Ferguson, 1999; Prachartam et al., 1996].

The five cephalometric variables (SNA, SNB, NSBa, NL-NSL, ML-NSL) used in the study on OSAS preschool children are able to describe the craniofacial morphology assessing the general pattern of craniofacial association [Solow, 1966; Hasund, 1977]; the facial type is mainly classified into retrognathic, orthognathic, and prognathic according to the relation of the maxilla and mandible to the anterior cranial base.

Significant correlations among the cephalometric values of the jaws on the sagittal (SNA, SNB), vertical plane (NL-NSL, ML-NSL) and the cranial base angle (NSBa) have been shown and clinically it is possible to describe the harmonious combination of these basal variables in a floating norms system dependent upon the type of the face [Hasund, 1977]. The floating norms concept was introduced by Hasund and substituted the "norm" intended as a population mean with a new norm constructed on the association among five correlated craniofacial measurements (SNA, SNB, NL-NSL, ML-NSL, NSBa). This concept has been applied only for adult subjects; Tollaro et al. provided the floating norms systems to assess skeletal pattern in subject with full deciduous and mixed dentition as guidance for early diagnosis and treatment planning [Tollaro et al., 1996].

The combinations of the cephalometric variables for retrognathic, orthognathic and prognathic face type are given in a graphical box-like form (the cephalometric box of floating norms). The basal configuration of the face can be verified plotting the individual measurements in the cephalometric box and drawing a connecting line among them; a straight line in the template framed by the tolerance limits indicates an harmonious relationship, a broken line not framed from the tolerance limits shows a disharmonious craniofacial morphology.

In the present study, according to the floating norms box for deciduous dentition, the OSAS preschool children showed an orthognathic and harmonious maxilla in the sagittal (SNA) and vertical plane (NL-NSL) in relation to the anterior cranial base.

The cephalometric values that are disharmonious with the orthognathic facial pattern regard the mandible. The angles SNB and ML-NSL lie outside the template of harmony with retrognathic range, showing a slightly retropositioned mandible with a strong posterior inclination in clockwise direction, in relation to the anterior cranial base. This skeletal disharmony causes a mild disto-basal sagittal relationship between the jaws but a severe increase of the facial height. The mild chin deficiency and the strong posterior mandibular inclination can also be seen in the Wiggle diagram when the OSAS sample is compared to an ideal occlusion group with deciduous dentition. These findings are in agreement with the results of other authors [Battagel et al., 2000; Cozza et al., 2004; Hochban et al., 1994; Kulnis et al., 2000; Linder Aronson, 1975; Under-Aronson and Woodside, 2000; Löfstrand-Tideström et al., 1999; Özdemir et al., 2004; Zuconi et al., 2009].

**Table 1** - Mean and standard deviation of the cephalometric variables of:

<table>
<thead>
<tr>
<th>Variables</th>
<th>OSAS subjects</th>
<th>Mean</th>
<th>SD</th>
</tr>
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<tbody>
<tr>
<td>Age (years)</td>
<td>4.57</td>
<td>0.59</td>
<td></td>
</tr>
<tr>
<td>SNA</td>
<td>79.63</td>
<td>2.64</td>
<td></td>
</tr>
<tr>
<td>SNB</td>
<td>74.00</td>
<td>3.08</td>
<td></td>
</tr>
<tr>
<td>ANB</td>
<td>5.55</td>
<td>1.86</td>
<td></td>
</tr>
<tr>
<td>NSBa</td>
<td>132.32</td>
<td>5.64</td>
<td></td>
</tr>
<tr>
<td>NL-NSL</td>
<td>39.11</td>
<td>4.76</td>
<td></td>
</tr>
<tr>
<td>ML-NSL</td>
<td>6.71</td>
<td>2.04</td>
<td></td>
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</table>

<table>
<thead>
<tr>
<th>Variables</th>
<th>Tollaro’s sample</th>
<th>Mean</th>
<th>SD</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td>5.67</td>
<td>0.82</td>
<td></td>
</tr>
<tr>
<td>SNA</td>
<td>79.88</td>
<td>2.91</td>
<td></td>
</tr>
<tr>
<td>SNB</td>
<td>76.35</td>
<td>2.85</td>
<td></td>
</tr>
<tr>
<td>ANB</td>
<td>3.53</td>
<td>2.63</td>
<td></td>
</tr>
<tr>
<td>NSBa</td>
<td>131.56</td>
<td>4.44</td>
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</tr>
<tr>
<td>ML-NSL</td>
<td>35.23</td>
<td>3.27</td>
<td></td>
</tr>
<tr>
<td>NL-NSL</td>
<td>7.94</td>
<td>2.75</td>
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at increasing the airflow to normalise breathing and to lower facial height. Mandibular plane and increased skeletal divergency of the maxilla and a mild retrognathic mandible with a steep deciduous dentition have an orthognathic, harmonious growth pattern.

**References**


