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Cervical column morphology in adult patients with obstructive sleep apnoea

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SUMMARY Cervical column morphology was examined in adult patients with obstructive sleep apnoea (OSA) and compared with the cervical morphology of an adult control group with neutral occlusion, normal craniofacial morphology, and no history of sleep apnoea. The sleep apnoea group consisted of 91 patients, 16 females aged 29–59 years (mean 49.4 years) and 75 males aged 27–65 years (mean 49.0 years). All patients were diagnosed with OSA by overnight polysomnography. The control group consisted of 21 subjects, 15 females aged 23–40 years (mean 29.2 years) and 6 males aged 25–44 years (mean 32.8 years). From each individual, a visual assessment of the cervical column was performed on the radiograph. Differences in the cervical column morphology, between the genders and the groups were assessed by Fisher's exact test and the effect of age by logistic regression analysis.

In the OSA group, 46.2 per cent had fusion anomalies of the cervical column and 5.5 per cent a posterior arch deficiency. Fusion anomalies occurred in 26.4 per cent as fusions between two cervical vertebrae. Block fusions occurred in 12.1 per cent and occipitalization in 14.3 per cent. A posterior arch deficiency occurred in 2.2 per cent as a partial cleft of C1 and in 3.3 per cent as dehiscence of C3 and C4. No statistical gender differences were found in the occurrence of morphological characteristics of the cervical column. The fusion anomalies of the cervical column occurred significantly more often in the OSA group.

The results indicate that the morphological deviations of the upper cervical vertebrae play a role in the phenotypical subdivision and diagnosis of OSA.

Introduction

Guilleminault *et al.* (1976) first described sleep apnoea syndrome. The word apnoea is derived from Greek and means without breathing. There are three types of apnoea: obstructive, central, and mixed. Obstructive apnoea is defined as cessation of airflow with persistent respiratory effort, due to collapse of the upper airways. Central apnoea is a cessation of airflow with no respiratory effort due to a short withdrawal of the central nervous drive to the respiratory muscles. Mixed apnoea is central apnoea passing into obstructive apnoea. In addition to the three described types of apnoea, there are hypopnoeas that are just as injurious as apnoeas. Obstructive hypopnoea is reduction in airflow with persistent respiratory effort, similar to all types of apnoea, lasting at least 10 seconds, followed by oxygen desaturation.

Obstructive sleep apnoea (OSA) is characterized by loud snoring, arousals, sleep fragmentation, intermittent hypoxaemia, and daytime sleepiness (e.g. Guilleminault *et al.*, 1976, 2006; Prisant *et al.*, 2006). OSA is by far the most common sleep-related breathing disorder, affecting 2–4 per cent of the adult population, particularly males aged 60 years and older where the prevalence is 30–60 per cent (e.g. Stradling and Crosby, 1991; Prisant *et al.*, 2006; Wolkove *et al.*, 2007).

OSA is multifactorial with age, gender, and body mass index (BMI) as predisposing factors (e.g. Guilleminault *et al.*, 1976; Paoli *et al.*, 2001; Prisant *et al.*, 2006). Most authors agree that there are craniofacial morphological and postural characteristics in patients with OSA such as a reduced posterior airway space, an abnormally long soft palate, a low position of the hyoid bone, and an extended head posture (Lowe *et al.*, 1986, 1996; Solow *et al.*, 1993, 1996; Petri *et al.*, 1994; Tangugsorn *et al.*, 1995; Pracharktam *et al.*, 1996; Paoli *et al.*, 2001; Hoekema *et al.*, 2003; Wong *et al.*, 2005).

Recently, an association between the morphology of the cervical column and craniofacial morphology and head posture has been demonstrated (Sonnesen and Kjær, 2007a,b; Sonnesen *et al.*, 2007). An association between fusion of the bodies in the cervical column as well as deviations in the vertical and sagittal craniofacial morphology was found (Sonnesen and Kjær, 2007a,b). Additionally, an association between fusion of the bodies in the cervical column and posture of the head and neck was observed (Sonnesen *et al.*, 2007).

The aetiology of OSA is still not fully understood. As age, gender, BMI, specific types of craniofacial morphology, and head posture are factors associated with OSA, fusion of the bodies in the cervical column in OSA patients could also be an associated factor.

Therefore, the aims of the present study were to describe the morphology of the cervical column in adult patients with OSA and to compare the morphology of the cervical column in a group of adult OSA patients with subjects with

neutral occlusion, normal craniofacial morphology, and no history of OSA (control group).

Subjects

The OSA group consisted of 91 patients, 16 females aged 29–59 years (mean 49.4 years) and 75 males aged 27–65 years (mean 49.0 years). The patients were all treated at the Department of Orthodontics, School of Dentistry, Copenhagen, Denmark, in the years 1999–2003. They were diagnosed with OSA by sleep studies, using overnight polysomnography. Only patients with the five or more apnoeas and hypopnoeas per hour of sleep [the apnoea–hypopnoea index (AHI)] were included in the study. AHI ranged between 5.1 and 111.7 (mean 36.0) and the BMI between 21.0 and 52.0 (mean 31.4). The mean horizontal overjet was 4.2 mm and the mean vertical overbite 2.6 mm.

The control group consisted of 21 subjects, 15 females aged 23–40 years (mean 29.2 years) and six males aged 25–44 years (mean 32.8 years). The subjects were either students or staff members at the School of Dentistry, Århus, Denmark. The selection criteria were the following: (1) neutral occlusion or minor malocclusion not requiring orthodontic treatment according to the Danish procedure for screening the population for malocclusions entailing health risks (Danish Ministry of Health, 1990; Solow, 1995); (2) no previous history of orthodontic treatment; (3) a sagittal and vertical jaw relationship within one standard deviation according to the standard material described by Björk (1947) assessed on lateral radiographs of each individual; (4) at least 24 permanent teeth present; (5) no craniofacial anomalies, systemic muscle, joint disorders, or history of sleep apnoea; and (6) accessibility of a profile radiograph before orthodontic treatment with the five first cervical vertebrae units visible. The control group has previously been described in detail (Sonnesen *et al.*, 2007).

Methods

Morphology of the cervical vertebrae

The visual assessment of the cervical column consisted of the first five cervical vertebral units that are normally seen on a standardized lateral skull radiograph. Characteristics of the cervical column were classified according to Sandham (1986) and divided into two categories: 'posterior arch deficiency' and 'fusion anomalies'. Posterior arch deficiency consisted of partial cleft and dehiscence and fusion anomalies of fusion, block fusion, and occipitalization.

Lateral profile radiographs

For the control group, the profile radiographs were taken with the teeth in occlusion and in the standardized head position, the mirror position, as described by Siersbæk-Nielsen and Solow (1982). The radiographs were taken at the Department of Oral Radiology, School of Dentistry, Århus, Denmark, in a Hofman Selectomat with a film-to-focus distance of 180 cm and a film-to-median plane distance of 10 cm. No correction was made for the constant linear enlargement of 5.6 per cent. A plumb line was hung from the ceiling to mark the true vertical line on the radiographs. The digital radiographic system was a photostimulable phosphor plate, Digora (Soredex, Helsinki, Finland) placed in a traditional cassette without an intensifying screen.

For the OSA group, the profile radiographs were taken at the Department of Orthodontics, School of Dentistry, Copenhagen, Denmark, in a Tagarno Philips/Valmet BR 2002 Cephalix cephalostat with a film-to-focus distance of 195 cm and a film-to-median plane distance of 15 cm. No correction was made for the constant linear enlargement of 8.3 per cent. An aluminium wedge, placed between the cassette and the patient's face, and a movable grid were used to increase the sharpness of the image.

Reliability

The reliability of the visual assessment of the morphological characteristics of the cervical vertebrae was determined by interobserver evaluations between the authors, showed very good agreement ($\kappa = 0.82$) as assessed by the kappa coefficient (Cohen, 1960).

Statistical analysis

For the occurrence of morphological characteristics of the cervical column, the differences between the genders and the groups were assessed by Fisher's exact test and for the effect of age by logistic regression analysis. The results of the tests were considered to be significant at $P < 0.05$. Statistical analyses were performed using the Statistical Package for Social Sciences, version 13.00 (SPSS, Inc., Chicago, Illinois, USA).

Results

In the OSA group, 46.2 per cent had fusion anomalies of the cervical column and 5.5 per cent posterior arch deficiency (Table 1). Fusion anomalies occurred in 26.4 per cent as fusions between C2 and C3, C3 and C4, or C4 and C5. Block fusions occurred in 12.1 per cent as fusions between C2, C3, and C4, C2, C3, C4, and C5, or C3, C4, and C5 (Figure 1). Occipitalization occurred in 14.3 per cent in combination with fusions, block fusions, or as a single deviation. A posterior arch deficiency occurred in 2.2 per cent as a partial cleft of C1 and in 3.3 per cent as dehiscence of C3 and C4 (Figure 2). No statistical gender differences or effect of age were found in the occurrence of morphological characteristics of the cervical column (females 62.5 per cent, and males 42.7 per cent).

In the control group, 14.3 per cent had fusion of the cervical column and 4.8 per cent both fusion and posterior arch deficiency (Table 1). The fusions always occurred between C2 and C3. No block fusions or occipitalizations were found in the control group. Only partial clefting of C1 was found in the control group. No statistical gender differences or effect of age were found in the occurrence of morphological characteristics of the cervical column (females 13.3 per cent, males 16.7 per cent).

Comparison of the OSA group with the control group showed that fusion anomalies of the cervical column occurred significantly more often in the OSA group ($P < 0.01$, Table 1).

Table 1 Prevalence of morphological characteristics of cervical column in patients with obstructive sleep apnoea (OSA) and in subjects with neutral occlusion and normal craniofacial morphology (control group).

Variable	OSA		Control group		*P*
	n	%	*n*	%	
Normal	58	52.7	18	85.7	**
Fusion anomalies	42	46.2	3	14.3	**
Fusion	24	26.4	3	14.3	NS
Block fusion	11	12.1	0	0.0	NS
Occipitalization	13	14.3	0	0.0	NS
Posterior arch deficiency	5	5.5	1	4.8	NS
Partial cleft	2	2.2	1	4.8	NS
Dehiscence	3	3.3	0	0.0	NS
More than one deviation	9	9.9	1	4.8	NS

**$P < 0.01$; NS, not significant; Fisher's exact test.

Discussion

In the present study, cervical column morphology was examined in adult patients with OSA and compared with the cervical column morphology of an adult control group with neutral occlusion and normal craniofacial morphology. A similar comparison has not previously been reported in the literature.

Figure 1 Morphological characteristics of the cervical column in patients with obstructive sleep apnoea. B: Block fusions of the cervical vertebral bodies (fusion anomalies).

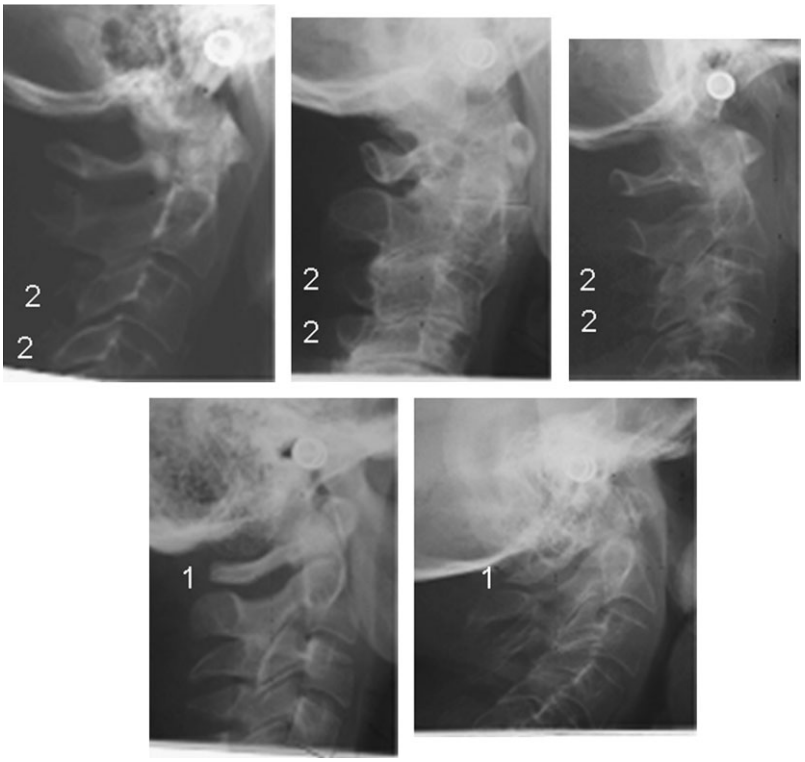

Figure 2 1: Partial cleft of the posterior portion of the neural arch of atlas (posterior arch deficiency). 2: Dehiscence of the third and forth cervical vertebral arches (posterior arch deficiency).

Although the control group was small, the study provides useful and valid information. It was not possible to extend the control sample as it consisted of healthy subjects with no medical indications for radiology. Therefore, profile radiographs of such a group are difficult to obtain in Denmark due to Danish legislation implemented by the national ethical committee.

In the sleep apnoea group, as well as in the control group, no statistical age or gender effect was found regarding cervical column morphology. Therefore, the control group was considered sufficient even though there were differences in age and gender between the two groups.

The morphological deviations of the cervical column vertebrae occurred significantly more often in the OSA group (46.2 per cent) compared with the control group, but the prevalence was approximately the same as in a group of patients with a skeletal deep bite (41.5 per cent; Sonnesen and Kjær, 2007a). On the other hand, the prevalence of the morphological deviations of the cervical column was larger in a group of patients with a skeletal mandibular overjet (61.4 per cent; Sonnesen and Kjær, 2007b) and in subjects with condylar hypoplasia (72.7 per cent; Sonnesen *et al.*, 2007).

The fusions of the cervical vertebral bodies seen in the controls always occurred between C2 and C3, while these in the OSA group seemed to be located more caudally in the cervical column as the fusions occurred between C2 and C3, C3 and C4, or C4 and C5. Furthermore, the pattern of posterior arch deficiency was different in the OSA group. Thus, in the control group, the posterior arch deficiency always occurred as a partial cleft of C1, whereas in the OSA group, the posterior arch deficiency also occurred as dehiscence of C3 and C4.

These interesting deviations in prevalence and pattern in the cervical column could not be explained in the present study, but the observations may prove to be a factor in the pathogenetic background for sleep apnoea and thereby contribute to the diagnosis, subdivision, and treatment of patients with OSA.

So far, the complex aetiology of OSA is still not fully understood, but a number of studies have previously contributed to phenotypic differentiations between types of sleep apnoea. It is well known that older males with an increased BMI are at risk of developing OSA (e.g. Stradling and Crosby, 1991; Prisant *et al.*, 2006; Wolkove *et al.*, 2007). Furthermore, specific types of craniofacial morphology and head posture such as a reduced posterior airway space, an abnormally long soft palate, a low position of the hyoid bone, and an extended head posture (Lowe *et al.*, 1986, 1996; Solow *et al.*, 1993, 1996; Petri *et al.*, 1994; Tangugsorn *et al.*, 1995; Pracharktam *et al.*, 1996; Paoli *et al.*, 2001; Hoekema *et al.*, 2003; Wong *et al.*, 2005) are also considered predisposing factors. In a previous study, an association was found between the posture of the

head and neck and fusions of the upper cervical vertebrae (Sonnesen *et al.*, 2007). As posture of the head and neck is considered to be associated with OSA, it is possible that OSA is associated with fusions of the cervical column.

In several familial and twin studies, it has also been shown that the aetiology of OSA is strongly influenced by genetic factors (Strohl *et al.*, 1978; Guilleminault *et al.*, 1995; Pillar and Lavie, 1995; Redline *et al.*, 1995; Gislason *et al.*, 2002; Kaparianos *et al.*, 2006).

The findings of the present study indicate a possible method to differentiate phenotypically between inherited and acquired OSA. If fusion of the cervical column occurs in patients with OSA, it could indicate that OSA was inherited. In the future, this may guide the clinician to choose between surgical and non-surgical treatment procedures and possibly allow prediction of the treatment outcome.

Conclusions

In the OSA group, 46.2 per cent had fusion anomalies of the cervical column and 5.5 per cent a posterior arch deficiency. Fusion anomalies occurred in 26.4 per cent as fusions, block fusions in 12.1 per cent, and occipitalization in 14.3 per cent. A posterior arch deficiency occurred in 2.2 per cent as a partial cleft of C1 and in 3.3 per cent as dehiscence of C3 and C4. Fusion anomalies of the cervical column occurred significantly more often in the OSA group. Not only was the prevalence of anomalies of the cervical column different from the control group but the pattern of anomalies of the cervical column was also different in the OSA subjects compared with the controls.

As these malformations in the cervical column have not previously been described, the results may prove to be important with regard to phenotypical subdivision, diagnosis, and treatment of OSA.

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