An investigation into the relationship between the severity of obstructive sleep apnoea/hypopnoea syndrome and the vertical position of the hyoid bone

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Objective: This study aimed to identify a correlation of diagnostic clinical significance between the vertical position of the hyoid bone in relation to structures within the dentofacial skeleton and:

(1) The severity of obstructive sleep apnoea hypopnoea syndrome, (OSAHS),
(2) Decisions relating to the management of the condition.

Design and Setting: Randomised retrospective survey of cephalometric records of subjects having been diagnosed as suffering with OSAHS by in patient overnight polysomnographic testing at the Edinburgh Royal Infirmary Sleep Centre, 2001-2002. **Materials and Methods:** Pre-polysomnograph orthoposition lateral cephalograms of 94 subjects tested, during the period from April 1996 to September 1997, were randomly selected and traced following strict adherence to standard protocol. Edentulous arches (one or both) formed the only exclusion criterion owing to obvious effects upon vertical dimensions of the cervico-pharyngeal region. Measurement of the vertical position of the hyoid bone was made relative to a number of planes validated by numerous previous cephalometric investigations, and these were recorded along with the apnoea/hypopnoea index (AHI), and subsequent management (mandibular repositioning appliances (MRA) / continuous positive airway pressure (CPAP)). Correlations between measurements and AHI were investigated using Spearman's Correlation Coefficients, and analysis of the relationship between hyoid bone position and management groups was undertaken using Wilcoxon Ranked Sum Testing. **Discussion and Results:** Statistically significant correlations were found between all linear measurements locating the hyoid bone in the vertical plane and subject AHI. The linear relationships were less reliable for subjects with AHI > 100, possibly due to a breakdown in the body's ability to respond posturally in order to maintain airway patency in more extreme cases. When the treatment groups (MRA/CPAP) were considered independently there was found to be a clear delineation between the two groups at a length of 120mm between the sella (S- a point upon the anterior cranial base) and the hyoid (H). This, in turn, may suggest that cephalometric radiographs may be used as a reproducible diagnostic tool

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INTRODUCTION

The alleged 'gold' standard investigation for the diagnosis of the sleep apnoea/hypopnoea syndrome (OSAHS) is inpatient overnight multichannel polysomnography. However, the ever-increasing demand for diagnosis of OSAHS has begun to place a strain upon services based on overnight laboratory studies. This situation has been clearly reflected at the Edinburgh Sleep Clinic where new referrals for polysomnographic testing were found to have increased ten fold between 1990 and 1996.[1] Whilst the diagnostic superiority of polysomnography is without question, it is accepted as being both time consuming and expensive. One recent report estimates the cost of such exhaustive diagnosis as approximating US$1,123 (~GB£700) per patient per night.[2] The introduction of devices suitable for home-based sleep studies has relieved some of the pressures associated with ever growing waiting lists. Investigations, however, have shown that as many as 56% of home-based studies require a further laboratory polysomnograph to ensure accurate diagnosis.[1] The recent literature investigating diagnostic alternatives reflects the understanding that traditional diagnostic services are becoming strained and that the management of subjects with mild to moderate forms of the condition may be delayed while awaiting exhaustive diagnosis that may not be essential.[2,3]

Consensus of opinion supports the concept of a heterogeneous aetiology of OSAHS and upper airway anatomy is often thought to play a significant role in the pathogenesis of the condition.[4,5] Indeed, data showing a familial aggregation of sleep disordered breathing strongly indicate that upper airway anatomy is an important determinant of upper airway closure in all patients with OSAHS.[6,7] Subjects typically present with a combination of cranio-mandibular abnormalities and it appears that one abnormality alone may not be sufficient to lead to the condition and that more complex relationships among cranio-mandibular elements may be involved. It is evident that the combination of a retro-positioned facial skeleton and reduced oropharyngeal dimensions, at one or more sites between the soft palate, tongue and pharyngeal wall, partly explains the aetiology of OSAHS. [4,8] Furthermore, the consensus of opinion indicates a threshold for its existence. That is, a number of predisposing factors may combine to exceed a threshold or balance between forces acting to maintain patency and those factors acting to oppose this task.

The hyoid bone would appear to provide a readily identifiable (radiographic) anatomic landmark, whose position is the result of a multifactorial postural system, which is closely aligned with maintaining respiratory function. Indeed, Djupesland *et al.* (1987) conclude that, "...with regard to skeletal morphology, the most outstanding feature in OSAHS subjects is the more inferior position of the hyoid bone".[9] Rather than being considered an anatomic aberration, a lower hyoid bone position, relative to the cranio-facial and cervical skeleton, might more accurately be deemed to reflect attempts made by the body's postural system to maintain airway patency. This patency may be compromised as a result of interactions between numerous factors of neurogenic, mechanical and anatomic origins.

MATERIALS AND METHODS

The records of ninety four subjects, who had undergone polysomnographic testing at the Edinburgh Royal Infirmary Sleep Clinic and subsequently found to have OSAHS, were randomly selected for assessment. The AHI range for the subject group was between 0 and 206, with a mean value of 46.4, and a distribution approximating that of the population examined within the sleep clinic; i.e. a greater number of subjects displaying relatively lower AHI values, as compared with extreme values of 40 and above. The subject group consisted of 74 males and 20 females, with an age range of 34 to 76 years inclusive, and a mean age of 54 years. Those subjects who were subsequently referred to the Victoria Hospital (Fife, Scotland) for treatment with mandibular repositioning appliances (MRAs) represented that portion of the investigated population suffering from a mild to moderate form of the OSAHS condition, a classification made following consideration of recordings of AHI, body mass index (BMI), EEG/EMG and associated medical status. In total, the investigation includes 57 subjects who have visited the Victoria Hospital for treatment with intra-oral appliances (MRAs), and 37 subjects who had been managed with alternative, less conservative measures.

Subjects who exhibited edentulous arches, (one or both), were excluded so as to eliminate inherent abnormalities associated with vertical face dimensions and consequently hyoid positions, but no effort was made to restrict the group to certain occlusal types. In an effort to obtain a sample truly representative of South East Scotland's general population at large, neither were subject to inclusion criteria restricted in terms of ethnicity, medical status, BMI, or subjective assessment results, (e.g. Epworth scale). There appears to be an imbalance between the sexes within the group and, while the ratio of men to women of approximately 4:1 is double that of 2:1 reported by other investigators.[10] The random nature of subject selection would indicate that this figure may be more representative of the subject population examined at the Edinburgh Sleep Clinic.

Cephalometric radiographs were obtained using a Siemens Orthophos CD at the Edinburgh Royal Infirmary Radiology Department. Images were recorded with the subjects standing upright with a 'natural' head position - the position of the head when standing with the visual axis being horizontal.[11]

Radiographs were traced to allow identification of specific anatomical points and planes, which in turn allowed appropriate measurements to be made. Tracings were performed in a darkened room on a well-illuminated viewing screen/tracing box using good quality acetate tracing paper and a 4H pencil. Ten hard tissue points were identified (Table 1), which allowed the plotting of the five reference planes necessary for the measurements required, (Table 2 and Figure 1). All measurements were adjusted for magnification factors.

ERROR CALCULATIONS

From the investigated population group, 25 radiographs were randomly selected and both the tracings and subsequent measurements were repeated for each case. This was undertaken by the same examiner with a minimum interval of a week between initial and repeat tracings. Analysis of the initial and repeated measurements is outlined in Table 3 and 4. In an effort to minimise the impact of systematic error, patients were selected from clinic records in order of their presentation for assessment/date of radiography, from April 1996. Only edentulous candidates and four others found to have radiographs unsuitable for this investigation were excluded, (in all four dentate cases which were excluded, point CV4ip was not visible). Tracings were performed only after the 'masking' of identifying markings such as the name and clinic number of the patient, so as to reduce the probability of subjective bias. All tracings and measurements were made by the author and adjusted for the measured degree of magnification.

Owing to the apparent distinction between variances within the groups (those who received treatment with MRAs and those who did not), the sample variance was taken only from the group identified as having received MRA treatment. The sample variances of this group were found to be lower, as compared with that of the entire investigated population and it was felt that the existence of random errors within the lower AHI population might be underestimated if this distinction was not made.

All data were analysed using SAS Professional Package Version 8. Evaluation of frequency histograms revealed that all variables excepting age, displayed a 'non – normal' distribution and hence non-parametric statistical procedures were utilised for data analysis. In the first instance, the relationships existing between AHI and the measurements pertaining to the position of the hyoid bone (H) relative to the third cervical vertebra and to the anterior cranial base (S) were analysed to determine the Spearman's correlation coefficients. All tests were performed at the 95% level of significance.

When the scatter plot of AHI versus S-H, involving subjects having been classified as having OSAHS of mild to moderate severity (i.e., those referred for MRA treatment), as against those deemed too severe for such treatment, was investigated there was an apparent distinction. This was subjected to Wilcoxon two sample tests for both AHI and S-H.

RESULTS

All linear measurements relating to hyoid bone position displayed a statistically significant correlation with increasing AHI (i.e. $p<0.05$, CI does not include 0), (Table 5).

The observation of apparent differences, visible within the plot of S-H vs. AHI, between the subjects who had been referred for management with MRA constructed at the Victoria Hospital and those who had been managed by other means via the Sleep Clinic, indicated a possible threshold of S-H distance (mm) with diagnostic potential, (Figure 2).

The plot indicates the existence of a threshold S-H

TABLE 1. RADIOGRAPHY IDENTIFIABLE ANATOMIC LANDMARK POINTS UTILISED IN THIS INVESTIGATION

Anatomic point	Description
Sella (S)	Sella turcica (mid point of the sella turcica)
Nasion (N)	Nasion (most anterior point on the frontonasal suture)
Orbitale (Or)	Orbitale (most inferior anterior point on the orbital margin)
Porion (Po)	Porion (uppermost, outermost point on the bony external auditory meatus)
Retrognathion (RGn)	Retrognathion (posterior most point upon the posterior surface of mandibular symphysis)
Hyoidius (H)	Hyoidius (most anterior and superior point of the hyoid bone)
CV2tg	The tangent point, on the dorsal contour of the odontoid process of the second cervical vertebra, to a line from CV2ip
CV2ip	Most posterior-inferior point of the corpus of the second cervical vertebra
CV4ip	Most posterior-inferior point of the corpus of the fourth vertical vertebra
C3	Most anterior-inferior point of the corpus of the third cervical vertebra

TABLE 2. RADIOGRAPHY IDENTIFIABLE ANATOMIC PLANES AND ANGLES UTILISED IN THIS INVESTIGATION

Plane/Angle/ Distance (mm)	Definition
FrP	Frankfort Plane/ Frankfort Horizontal (line joining Orbitale with Porion)
SN	A line joining Nasion with Sella turcica
C3-FrP	Line running parallel to the FrP, from point C3
C3-SN	Line running parallel to SN, from point C3
C3-RGn	Line running from C3 to RGn
OPT	Odontoid process tangent, through CV2ip and CV2tg
CVT	Cervical vertebra tangent, through CV4ip and CV2tg
OPT/CVT	Angle between the odontoid process and the cervical vertebra tangent, (conventionally used as an indication of flexure of the cervical vertebral column)
(C3-Frp) H	Distance measured from point H along the perpendicular to line C3-Frp
C3-SN - H	Distance measured from point H along the perpendicular to line C3-SN
S-H	Distance (straight line) between point S and point H

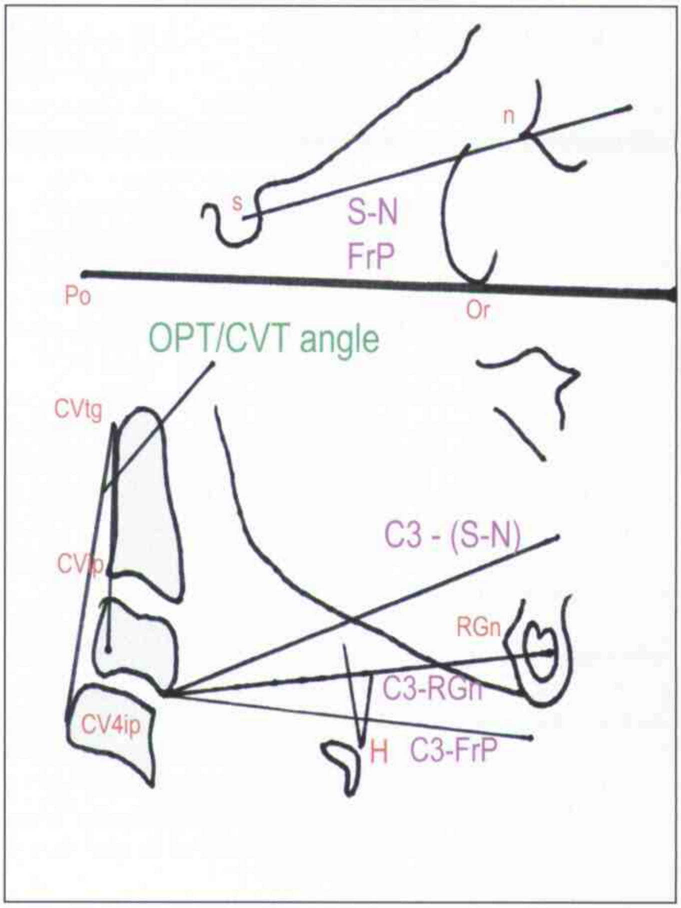

***Figure 1:** Cephalometric points and planes utilised in the analysis of this investigation*

value of 120mm. Taking the assumption that the decision relating to a subject's suitability for MRA management or otherwise, while based primarily upon AHI/OSAHS severity, is influenced by a number of factors including EMG activity results, expected CPAP compliance, and associated medical conditions. A diagnostic value of S-H = 120mm could be said to be significant in terms of treatment decisions. Namely, SH< 120mm is MRA management suitable and SH> 120mm is MRA management unsuitable.

Wilcoxon two sample rank sum testing of AHI and S-H variables between the two subject groups confirmed the statistically significant differences in both groups, ($p<0.0001$).

DISCUSSION

The results of this investigation demonstrate statistically significant correlations between the severity of OSAHS (AHI) and all linear measurements relating the hyoid bone to associated anatomic planes and points within the vertical plane. This confirms earlier findings, which indicate the existence of a correlation between the severity of obstructive sleep apnoea and vertical hyoid bone position.[12,13] With regard to those measurements made from planes arising from point C3, the distance of the hyoid (point H) along the perpendicular from the C3-retrognathian plane appeared to show the strongest

TABLE 3. RESULTS OF SYSTEMATIC ERROR ASSESSMENT

	S-H	(C3-SN) - H	(C3-FrP) - H	(C3-RGn) - H	OPT/CVT
Systematic Error					
Mean difference	0.36	0.036	0.216	0.252	0.12
Root n	5	5	5	5	5
Standard deviation	8.8171	6.41521	7.92851	5.49254	4.40528
t	0.2041	0.02806	0.13622	0.2294	0.1362
Systematic Error indicated if t is greater than:					
5% significance	2.010634				
10% significance	1.677224				

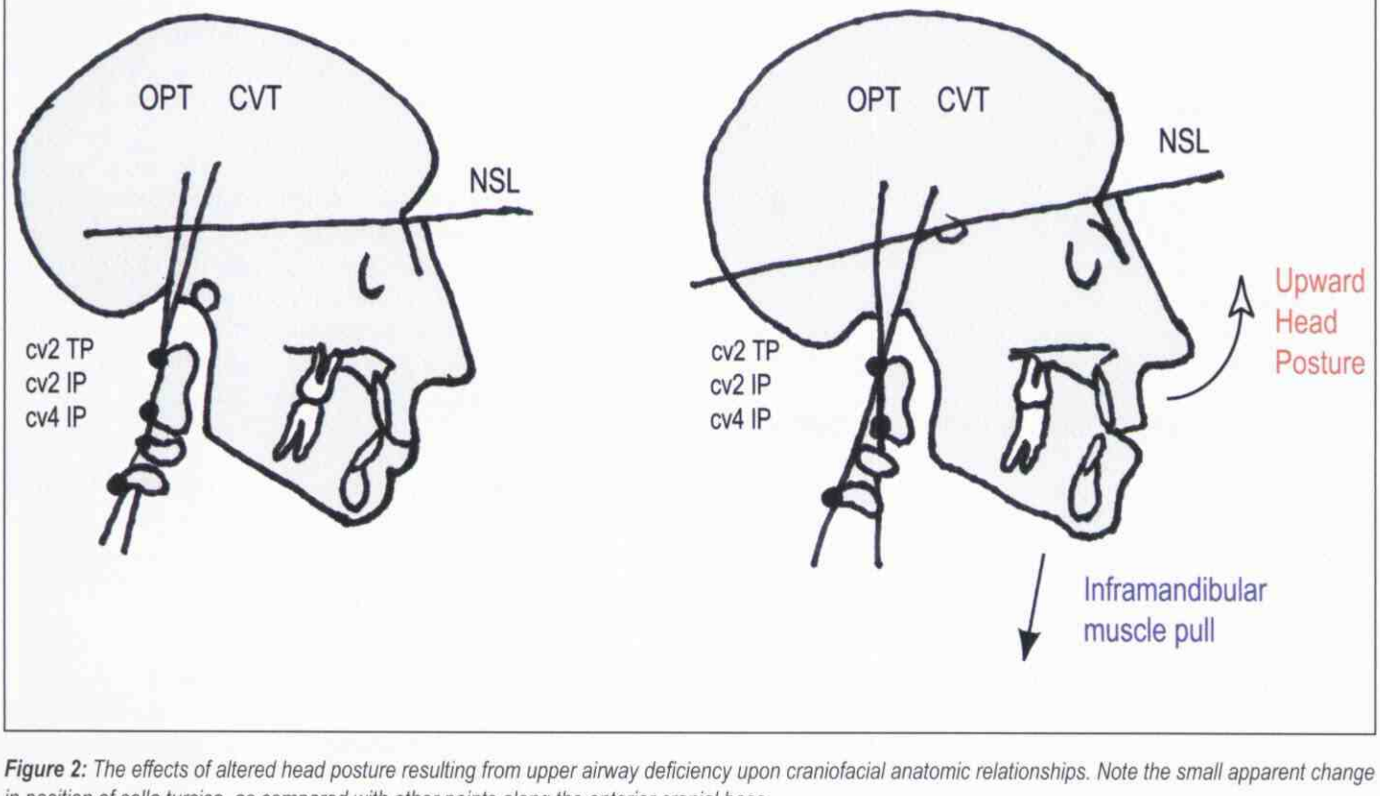

Figure 2: *The effects of altered head posture resulting from upper airway deficiency upon craniofacial anatomic relationships. Note the small apparent change in position of sella turcica, as compared with other points along the anterior cranial base.*

correlation (rs= -0.43, CI= -.058 to -.025, p< 0.0001) to AHI. Surprisingly, and apparently contrary to the findings of Bibby and Preston (1981), the linear measurement taken from anterior cranial base point S (sella turcica mid-point) directly to the hyoid (H) bone, demonstrates the strongest statistical correlation with AHI within the present subject group (rs= 0.72, CI= 0.61-0.81, p<0.0001). These results suggest that the inter-subject variability in hyoid bone position as related to the point S on the anterior cranial base, which one might attribute to postural differences and to expected differences in craniofacial dimensions (size), may be less significant than expected. Certainly, it would appear that the potential for variation for measurements taken within Bibby and Preston's more localised 'zone' appears more significant in the present investigation, as compared with that of direct measurement from the cranial base when one is considering a relationship with the severity of OSA. This may be explained by the fact that as the head tilts upwards and forwards (Figure 3), the position of point S within the skull might be affected to a lesser degree, as compared with other points upon the cranial base, and indeed the orientation and position of the cervical vertebrae. The latter effects may prove more significant as compared with difficulties one might experience when measuring hyoid position from a distant point, in different subjects of different craniofacial dimensions.

The relationship between hyoid bone position and postural adaptive changes aimed at maintaining airway patency appears to be almost universally accepted within the relevant literature.[14] Analysis of the relationship between hyoid bone position and OSAHS severity, as demonstrated within this investigation, irrespective of reference plane of measurement, is suggestive of an overloading of this adaptive system as AHI values approach high end values. All plots demonstrate a clear shift in the linear nature of the relationships between vertical hyoid position and AHI as the latter approaches and exceeds 100. This is thought to reflect a breakdown in the body's protective upper airway postural system in exceedingly severe forms of the OSAHS condition.

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TABLE 4. RESULTS OF RANDOM ERROR ASSESSMENT

	S-H	(C3-SN) - H	(C3-FrP) - H	(C3-RGn) - H	OPT/CVT
Random Error					
Error variance	0.6156	0.4374	0.9072	0.405	0.4
Sample variance	102.955	38.8957	48.2143	30.8949	11.9679
3% of sample variance	3.08867	1.166871	1.446429	0.926847	0.359037
10% of sample variance	10.2955	3.88957	4.82143	3.08949	1.19679
Intra-class Corr. Coeff (τ)	0.9935	0.9892	0.9864	0.9877	0.9826

TABLE 5. SPEARMAN'S CORRELATION TEST RESULTS FOR LINEAR MEASUREMENTS RELATING TO HYOID POSITION vs AHI				
	(C3-FrP) - H	(C3 - RGn) - H	(C3 - SN) - H	S-H
N	94	94	94	94
rs (Corr. Coeff)	-0.37	-0.43	-0.040	0.72
95% CI	-0.53 to -0.18	-0.58 to -0.25	-0.56 to -0.22	0.61 to 0.81
Two tailed p Approx and corrected for ties	<0.0011	<0.0001	<0.0001	<0.0001

SUMMARY

- **Statistically significant correlations have been identified between the vertical position of the hyoid bone and the severity (AHI) of the OSAHS condition. The vertical distance between the hyoid bone and the point S (sella turcica) upon the anterior cranial base demonstrates the highest correlation with OSAHS severity (AHI)**
- **Evidence suggests a possible overloading of the postural system associated with efforts to maintain upper airway maintenance (reflected by hyoid bone position) as OSAHS severity (AHI) reaches extreme values (> 100)**
- **A statistically significant difference is apparent at a sella to hyoid distance of approximately 120mm, between subjects who are deemed to suffer from a mild to moderate form of the OSAHS condition and are subsequently referred for MRA management (< 120mm), and those who are deemed to suffer from a severe form of the condition (>120mm). This linear measurement is proposed to be of clinical value in terms of its application in diagnostic triage and subsequent management decisions.**

The superiority of objective and complete multichannel polysomnographic testing is not questioned, and certainly in the presence of serious medical conditions thought to be associated with the OSAHS condition, it must be the diagnostic method of choice. There is, however, a need for simpler, cheaper diagnostic aids, certainly for those cases that exhibit simple snoring or mild OSAHS. The use of cephalometric radiographs to ascertain the sella - hyoid length, provides an extremely rapid and effective diagnostic tool, either used in isolation as a triage, or in combination with more intrusive diagnostic methods.