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Cervical vertebrae anomalies in orthodontic patients: a growth-based superimpositional approach

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SUMMARY The purpose of this study was to propose a growth-based structural superimposition method for assessment of cervical vertebral fusion and evaluate variations and abnormalities of the upper cervical vertebrae. Standardized lateral cephalograms of 156 patients (69 males and 87 females, age range 6–20 years), representing a skeletally heterogeneous orthodontic population, were used. Primary criterion for sample selection was the existence of at least two lateral cephalograms, one taken before orthodontic treatment, which depicted the first four cervical vertebrae. The abnormalities of the vertebrae were estimated by visual assessment and structural superimposition. Lateral cephalometric analysis was conducted in order to correlate vertebral anomalies to skeletal pattern. Descriptive statistics were calculated for all variables and interobserver agreement was evaluated using the kappa statistic.

Four patients (2.6 per cent) were found to have secondary ossicles in close relationship to the first cervical vertebra, while in 7.4 per cent, the vertebral arteries of the atlas were surrounded by a complete ring-shaped osseous structure. Three cephalograms showed atlas posterior arch dehiscence. After visual examination, 14 patients were provisionally identified as presenting fusion between the second and third cervical vertebrae. However, growth-based superimposition of the radiographs disclosed that no patient showed actual fusion, even though the lateral cephalometric analysis revealed sufficient extreme skeletal patterns, which have been previously related to vertebral fusion.

The findings of this study demonstrated a low percentage of atlas anomalies. It was not possible to correlate skeletal pattern to fusion of cervical vertebrae because no fusions were found. Subjective visual examination of a single cephalogram may result in false-positive findings of fusion and growth-based superimposition is recommended.

Introduction

Recently, there has been increased interest in the study of anomalies of the cervical vertebrae in the orthodontic literature, probably stemming from the demonstrated association between such anomalies and craniofacial syndromes (Gray *et al.*, 1964; Gunderson *et al.*, 1967; Brinker *et al.*, 1997; Guille and Sherk, 2002; Tracy *et al.*, 2004; Kaplan *et al.*, 2005), non-syndromic congenital anomalies, such as clefts (Ross and Lindsay, 1965; Sandham, 1986; Horswell, 1991; Uğar and Semb, 2000; Giannakari, 2004; Rajion *et al.*, 2006), and also conventional orthodontic malocclusions (Sonnesen and Kjær, 2007a,b, 2008a,b). Interstudy differences in the prevalence of these anomalies are large and difficult to explain; they could be attributed to true population differences or to methodological errors, arising from the choice of plain visual assessment as the method of evaluation. It is doubtful if this approach can be significantly improved because, besides being subjective and possibly unreliable, it is based on a single radiograph that has inherently limited information. However, integrating additional images, preferably at different postures or growth stages, could provide increased diagnostic confidence.

In orthodontics, the main concern is with the detection of the congenital subgroup of spine malformations, mainly fusions at the level of C2–C3 and atlas dehiscences, but in orthopaedics, where problems of the spine have been studied more extensively and in depth, fusions are even induced in order to correct spine deformities such as scoliosis (Mercado *et al.*, 2007). The specificity and sensitivity of various diagnostic methods to evaluate the success of such fusions is an active area of research. In addition to subjective observation of a single radiograph, more sophisticated methods have been investigated, including flexion–extension radiograph pairs (Taylor *et al.*, 2007), computed tomography (Brodsky *et al.*, 1991; Rajion *et al.*, 2006; Carreon *et al.*, 2007), and computer-aided quantitative motion analysis (Taylor *et al.*, 2007; Fassett *et al.*, 2008). Such procedures are not indicated for routine orthodontic patients because congenital fusions of the cervical spine are low in frequency and do not present with significant clinical manifestations that require intervention (Klimo *et al.*, 2007). However, since incidental findings may be observed (Soni *et al.*, 2008), it would be beneficial to be able to arrive at a definitive diagnosis, based on the diagnostic records already available from orthodontic treatment.

The aims of the present study were (1) to propose a growth-based superimpositional approach for assessing cervical vertebral fusions, (2) to apply this method for recording the type and prevalence of upper cervical vertebrae anomalies in a skeletally heterogeneous orthodontic population, and (3) to assess any correlation between cervical spine anomalies and craniofacial skeletal pattern.

Materials

The sample consisted of standardized lateral cephalograms of 156 patients (69 males and 87 females), consecutively treated either in the Orthodontic Department, School of Dentistry, University of Athens, or in a private orthodontic practice. The lateral radiographs were selected irrespective of gender and type of malocclusion. Inclusion criteria were (1) age between 6 and 20 years, (2) at least two lateral cephalograms (one at the beginning of orthodontic treatment), and (3) radiographs of good quality, showing the first four cervical vertebrae and a reference ruler on the cephalostat, for exact measurement of the magnification factor. Exclusion criteria were previous orthodontic treatment and syndromes and other developmental deformities.

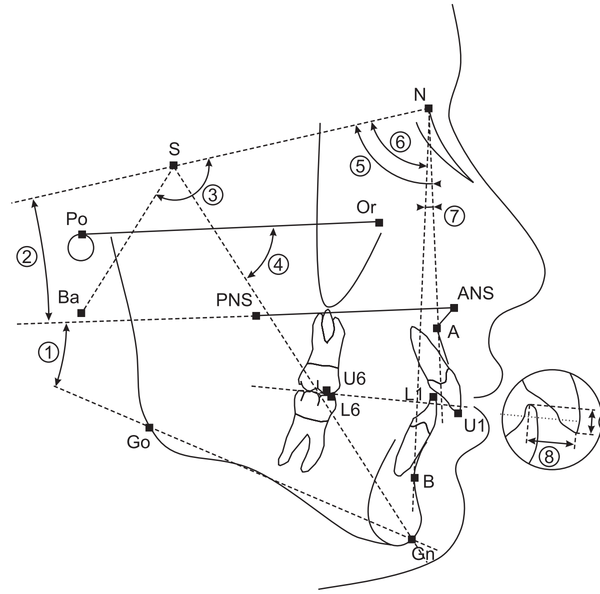

Figure 1 Points and measurements used for the cephalometric analysis. Porion (Po), orbitale (Or), sella (S), nasion (N), anterior nasal spine (ANS), posterior nasal spine (PNS), point A (A), point B (B), gnathion (Gn), gonion (Go), basion (Ba), upper incisor tip (U1), lower incisor tip (L1), lower molar mesial cusp (L6), and upper molar mesial cusp (U6).

Methods

All lateral radiographs were scanned at 150 dpi and saved as JPEG files.

Lateral cephalometric analysis

The initial radiographs were digitized by one author (DK) with Viewbox 3 software (dHal Software, Kifissia, Greece). A comprehensive cephalometric analysis was performed but only 15 skeletal and dental points were used in this investigation (Figure 1).

Nine variables which represented vertical and sagittal craniofacial dimensions were calculated. These were the angles ANS–PNS to Go–Gn, S–N to ANS–PNS, cranial base angle (N–S–Ba), Po–Or to S–Gn, SNA, SNB, ANB, and the linear measurements overjet and overbite.

To assess repeatability and measurement error, 20 radiographs were randomly selected, re-digitized, and re-measured by the same investigator, after a period of at least 1 week. Systematic error was assessed by paired *t*-tests (alpha = 5 per cent) and random error was evaluated according to Houston (1983). No systematic error was detected. Random error ranged from 0.28 to 0.86 degrees for angular measurements and from 0.52 to 0.57 mm for linear measurements. The reliability coefficient ranged from 92.5 to 99.0 per cent.

Cervical spine morphology

The radiographs were visually assessed by both authors separately and the prevalence of posterior cervical artery canal morphology, atlas dehiscence, accessory ossicles, and fusions were noted.

The first cervical vertebra was classified into four types according to the morphology of the posterior margin of the lateral articular processes, as described by Farman *et al.* (1979) and Farman and Escobar (1982): type 1, posterior margins of the atlas processes almost perpendicular to the posterior arch; type 2, superior arch of the processes form a short posterior lip; type 3, superior arch of the processes extends posteriorly but does not fuse with the posterior arch; and type 4, vertebral arteries surrounded by a complete ring-shaped structure.

A posterior arch dehiscence (rachischisis) of the atlas was recorded when a uniform radio-opacity without an internal cortical outline was observed at the distal margin of the posterior arch, signifying failure of fusion (Farman *et al.*, 1979; Farman and Escobar, 1982; Sandham, 1986; Jones, 1998).

Accessory ossicles were identified as independent radio-opaque structures in close relationship to the cervical units (Farman *et al.*, 1979; Farman and Escobar, 1982).

Fusions between the cervical vertebrae were identified as osseous continuities, without complete separation at the intervertebral disc or at the articular surfaces (Farman *et al.*, 1979; Farman and Escobar, 1982; Sandham, 1986).

Interobserver agreement was evaluated by comparing the individual results. Evaluations for atlas dehiscence were identical between the investigators. Assessment of the type of articular process of the atlas showed agreement in 70 per cent of the cases. When there was disagreement, it was only between neighbouring categories. The calculated weighted kappa statistic was 0.65 (Altman, 1991).

There was no agreement for fusions; one investigator identifying 14 fusion cases between the posterior arches or articular surfaces of C2 and C3, whereas the other examiner none. For further investigation, the disputed cases were traced and superimposed.

Structural superimposition

Tracing of C2 and C3 was performed according to the recommendations of Vastardis and Evans (1996), except for the area of disputed fusion which was left untraced. Three structural superimpositions of the tracings were carried out using a best-fit criterion, taking into account normal vertebral growth with enlargement occurring at the body–disc interface by physeal ossification (Dickson and Deacon, 1987): (1) superimposition of C2, (2) superimposition of C3, and (3) superimposition of the intervertebral disc space between C2 and C3. To avoid tracing errors, similar superimpositions were conducted using the original digital image files. These were processed by imaging software (Adobe Photoshop CS3, Adobe Systems Inc., San Jose, California, USA), first by automatic contrast adjustment and then by enhancement of the outlines of the osseous structures (glowing edges filter, colourizing with contrasting colours, and superimposition using a 'difference' blending mode).

If an osseous fusion between two vertebrae exists, then two consequences are expected: (1) the relative spatial relationship between the vertebrae should remain unchanged, even under extension or flexion of the spine and (2) growth of the vertebrae should result in characteristic features, not observed under normal circumstances. Growth of the vertebrae proceeds by endochondral ossification of the physeal plates that lie between the vertebral body and the intervertebral discs (Dickson and Deacon, 1987). The growth process is similar to that occurring in long bones, with the significant difference that there is no epiphysis beyond the physeal plates, which will fuse with the vertebral body. Thus, it is not possible to determine the age of growth cessation by radiographically observing the time of epiphyseal fusion. Studies of spinal elongation (Stokes and Windisch, 2006) have shown that increases in intervertebral disc height effectively cease after 10 years, but vertebral bodies continue growing even after 20 years of age. Thus, fusion of vertebrae at the posterior processes or at the articular surfaces during a period of active growth should result in progressive diminution of the intervertebral disc space and decreased overall spine length.

Results

Lateral cephalometric analysis and craniofacial dimensions

Descriptive statistics of the cephalometric measurements are shown in Table 1.

Cervical spine morphology

The percentage of each of the four types of morphology of the posterior margin of the lateral articular processes (Farman *et al.*, 1979; Farman and Escobar, 1982) was as follows: type 1, 33.0; type 2, 51.0; type 3, 8.7; and type 4, 7.4 (average values of the two investigators).

A midline dehiscence of the posterior arches of the atlas was observed in three cases (1.9 per cent). In all three, a uniform radio-opacity of the distal margins of the divided arch was observed, while in one patient, this was associated with a thin, boomerang-shaped profile of the vertebra (Figure 2).

Formation of accessory ossicles in the area between the atlas posterior arch and the base of the skull was observed in four patients (2.6 per cent).

Fourteen patients (9 per cent) were at first identified by one investigator as presenting fusion between the C2 and C3. All revealed a uniform radio-opacity in the area between the inferior articular surface of C2 and the superior articular

Table 1 Cephalometric analysis results. All measurements in degrees except where otherwise noted.

	Mean	Standard deviation	Range
Vertical			
ANS–PNS/Go–Gn	28.4	5.85	12.0 to 50.0
S–N/ANS–PNS	7.0	3.47	−0.2 to 18.3
S–N/Go–Gn	33.9	5.52	18.0 to 51.1
Po–Or/S–Gn	59.4	3.90	51.7 to 70.7
Sagittal			
SNA	80.0	3.84	70.4 to 89.7
SNB	76.3	3.91	66.1 to 88.7
ANB	3.7	3.21	−9.2 to 10.3
Cranial base			
N–S–Ba	131.4	7.80	60.1 to 144.4
Incisor relationship			
Overjet (mm)	5.2	3.23	−3.7 to 12.4
Overbite (mm)	2.4	2.39	−6.6 to 8.8

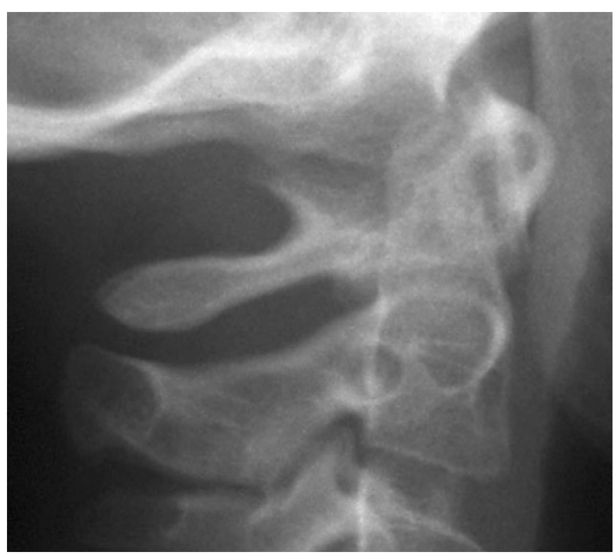

Figure 2 Patient no. 33. Atlas dehiscence.

surface of C3 on at least one of the radiographs. However, after tracing and superimposition of each of the 14 pairs of radiographs, no actual fusion was found. In all cases of suspected fusion, it was not possible to satisfactorily superimpose the complete C2–C3 structure (Figure 3). Separate superimpositions at C2 or C3 showed increased overall distancing between the two vertebrae with age, presumably an effect of unimpeded growth. The height of the intervertebral disc space remained relatively unchanged.

Relationship between facial morphology and fusion anomalies

Due to no fusions in the sample, it was not possible to arrive at any correlations between craniofacial morphology and fusion anomalies. However, since such relationships have been reported in the literature (Sonnesen and Kjær, 2007a,b, 2008a,b), a valid concern could be raised that perhaps the present sample did not encompass sufficiently 'extreme' facial types to produce a detectable number of fusions. Thus, the cephalometric results were used to calculate the expected fusions, based on the percentages reported by Sonnesen and Kjær (2007a,b, 2008a,b). The skeletal Class II category included 22 patients who had an ANB angle larger than 1 standard deviation (SD) from the average, whereas the skeletal Class III category included five patients. In the skeletal deep bite category, 24 patients were identified as having a S–N to Go–Gn angle more than 1 SD below the average; the corresponding number in the skeletal open bite category was 22. However, because of overlap with the Class II and Class III categories, these numbers were reduced to 20 and 13, respectively. Thus, by multiplying with the appropriate frequencies, the calculated total number of expected fusions was 43.

Discussion

The prevalence of cervical vertebrae anomalies has been reported in numerous studies in the literature but with a wide variation in results (Table 2). Explanations for this could include true interpopulation diversity, differences in methodological reliability, subjectivity, and lack of interobserver calibration.

Regarding fusions, two groups of studies are found, one reporting low prevalence, ranging from 0 to 4 per cent (Brown *et al.*, 1964; Ross and Lindsay, 1965; Sandham, 1986; Uğar and Semb, 2000; Giannakari, 2004; Rajion *et al.*, 2006), and the recent studies of Sonnesen and Kjær (2007a,b, 2008a,b) that quote values above 14 per cent,

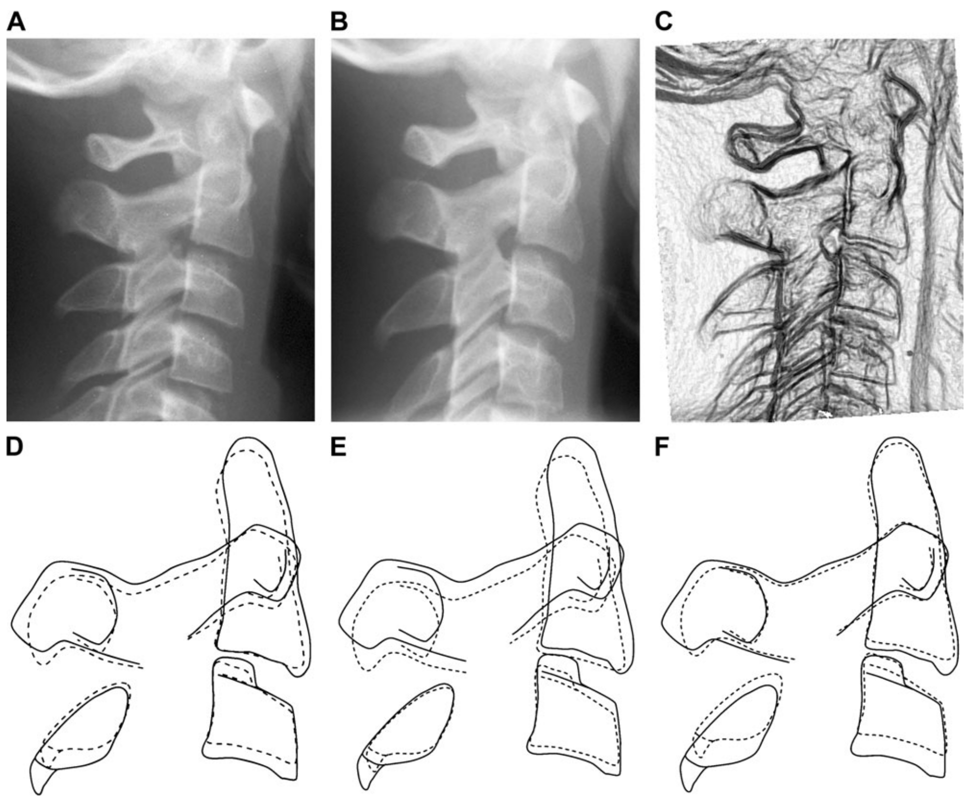

Figure 3 Patient no. 9 (male). (A) Initial radiograph at 13 years of age, (B) final radiograph 2 years later, (C) superimposition of the images at C2 after enhancement and colourization (see text; here reproduced in grey scale), and (D–F) tracing superimpositions at each of three areas: intervertebral disc space, C3, and C2, respectively. Note constant height of disc space and relative downward displacement of C3 when superimposing on C2 (C and F).

Table 2 Prevalence of cervical vertebrae anomalies as reported in the literature.

Authors	Year	Sample size	Accessory ossicles (%)	Vertebral artery type (%)	Atlas dehiscence (%)	Vertebral fusion (%)	Unspecified (%)
Brown *et al.*	1964	1400				0.71	
Ross and Lindsay	1965	800				0.75	
Farman *et al.*	1979	220	1.4	Type 1: 12.7 Type 2: 60.5 Type 3: 18.6 Type 4: 8.1	3.6		
Sandham	1986	120			0.8	0	
Horswell	1991	100					7
Uğar and Semb	2000	264			5	4.1	
Giannakari	2004	50	0		0	0	
Rajion *et al.*	2006	12			0	0	
Sonnesen and Kjær (skeletal deep bite group)	2007a	41			9.8	41.5	
Sonnesen and Kjær (skeletal Class III group)	2007b	57			12.3	61.4	
Sonnesen and Kjær (skeletal Class II group)	2008a	34			5.9	52.9	
Sonnesen and Kjær (skeletal open bite group)	2008b	38			13.2	42.1	
Sonnesen and Kjær (normal group)	2007a,b and 2008a,b	21			4.8	14.3	
Present study		156	2.6	Type 1: 33.0 Type 2: 51.0 Type 3: 8.7 Type 4: 7.4	1.9	0	

exceeding 60 per cent for skeletal Class III individuals (Table 2). The present results are in agreement with the first group, even though cephalometric analysis showed that the sample included a sufficient number of extreme skeletal patterns. On this basis, approximately 43 fusions could have been expected, a number significantly larger than even the 14 provisionally identified.

Most investigations in the orthodontic literature have used subjective visual examination of cephalometric radiographs. However, the criterion of absence of a continuous radiolucent area between the articular or spinous processes may not be valid because spine inclination, flexion or extension, and morphological variations could result in superposition of structures giving an analogous appearance. The gold standard for assessing fusions and other anomalies is direct observation on autopsy material or during surgical exploration (Brown *et al.*, 1964; Templeton and Brown, 1964; Brodsky *et al.*, 1991). No study could be identified that specifically assessed the sensitivity and specificity of cephalometric radiography in the cervical region, but studies evaluating post-surgical fusions in the lumbar region have shown low agreement between pre-operative radiographs and surgical results (Brodsky *et al.*, 1991; Blumenthal and Gill, 1993; Kant *et al.*, 1995).

Flexion–extension radiograph pairs are used extensively in the orthopaedic literature for evaluating post-surgical results of induced fusion (Brodsky *et al.*, 1991; Taylor *et al.*, 2007; Fassett *et al.*, 2008). Changes in intervertebral angulation can be assessed either by observation or by manual and computer-aided measurements (Taylor *et al.*, 2007; Fassett *et al.*, 2008). Subjective evaluation has low interobserver agreement, possibly due to lack of consensus criteria; quantitative measurements improve the results. However, flexion–extension pairs result in extra radiation dosage to the patient and this is not recommended for routine screening, considering that most fusions at the C2–C3 level remain asymptomatic and do not require any intervention (Klimo *et al.*, 2007). Even if two cephalometric radiographs, that happen to show significant differences in spine angulation, are available, measurements between the vertebrae are not expected to provide conclusive answers because the difference in angulation that can be achieved in a cephalostat is much smaller than that obtained from flexion–extension pairs taken in extreme spine angulation. In the present sample, the maximum spine angulation between C2 and C6 was approximately 20 degrees, whereas in diagnostic flexion–extension pairs, the values range from 31 to 80 degrees (Reitman *et al.*, 2004). Considering that the angulation at C2–C3 represents 15 per cent of the total (Reitman *et al.*, 2004), the maximum expected angulation between C2 and C3 in cephalometric radiography would be approximately 3 degrees, which may be difficult to measure. Furthermore, the presence of fusion does not imply zero angulation between the vertebrae because bone is inherently elastic and can exhibit deflection under stress (Bono *et al.*, 2007). There is

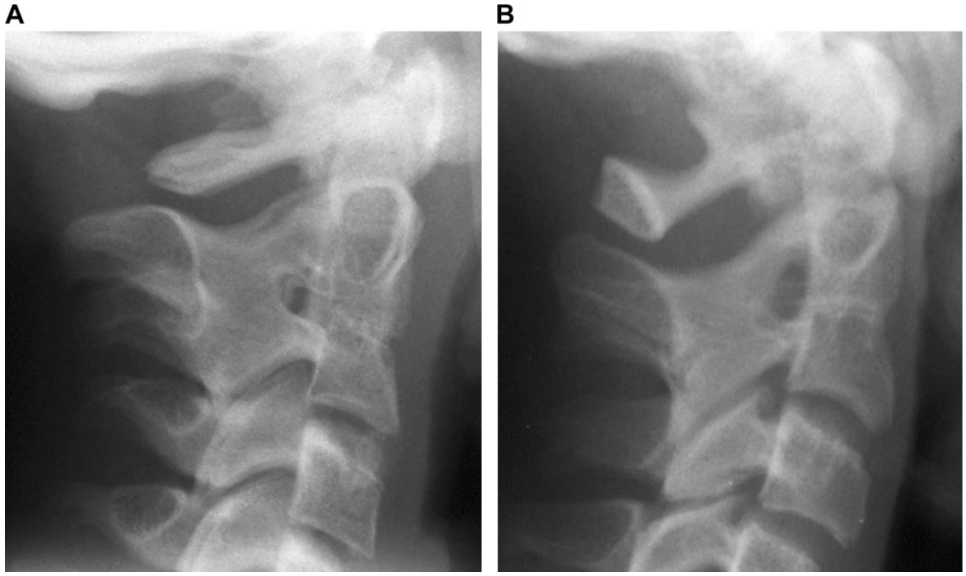

Figure 4 Obliteration of intervertebral disc space between C2 and C3 due to fusion of the articular processes. These patients were not part of the sample.

no consensus regarding the cut-off point signifying fusion but most investigators accept the value of 1.5 degrees (Taylor *et al.*, 2007; Fassett *et al.*, 2008). These factors make it unlikely that fusions could be detected with the small angular deviations seen between cephalometric radiographs.

A pre- and post-treatment orthodontic cephalogram are usually taken routinely during a period when there is active growth. Growth of the vertebrae occurs at the superior and inferior surfaces of the body in a similar manner to the epiphyseal plates of long bones (Dickson and Deacon, 1987). The intervertebral disc has been found to increase in size up to the age of approximately 10 years and remain constant thereafter, whereas the vertebral bodies increase in height beyond 20 years of age (Stokes and Windisch, 2006). In the presence of fusion, continuation of growth at the physeal plates is expected, thereby reducing or even obliterating the disc space (Brown *et al.*, 1964; Ritsilä and Alhopuro, 1975; Figure 4).

The present structural superimposition method is based on the intervening growth between the two successive cephalograms. The observed increase in vertebral body height confirms active growth, whereas separation of the vertebrae and the constant disc space confirm absence of fusion. In cases of suspected fusion at the initial examination, the time delay of 1 or 2 years is not a significant limitation, unless other signs point to syndromic conditions.

Conclusions

Assessment of fusions from a single cephalometric radiograph is highly subjective. A growth-based superimpositional method is proposed, that does not require extra radiation, beyond that used for routine orthodontic records.

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