Relationship between cervical and global sagittal balance in patients with dropped head syndrome

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Abstract

Background DHS is characterized by chin-on-chest deformity and devastatingly impedes activities of daily living in affected individuals. There is a paucity of literature about the pathophysiology of DHS including knowledge about spinal sagittal alignment. We conducted this study to clarify the relationship between cervical sagittal alignment and global sagittal balance in DHS.

Methods This is a retrospective radiographic study of a case series of DHS. Forty-one patients with diagnosed DHS were enrolled. Measurements were made using lateral standing radiograph.

Results C2–C7 sagittal vertical axis (SVA) was estimated as 52.0 ± 2.4 mm. Among sagittal parameters, C7–S1 SVA positively correlated with C2–C7 angle (C2–C7 A) ($r=0.33$). For the correlations between C7 and S1 SVA and C2–C7 A, both logistic and linear regression models were used to determine the threshold for C2–C7 A value responsible for global sagittal balance. C2–C7 A of -15.0 and 6.0 were predicted by logistic and linear regression models and were considered responsible for the occurrence of global positive imbalance. Therefore, we divided into two groups, namely, cervical kyphosis group (C type) and diffuse kyphosis group (D type) by median value of C2–C7 A. Enlarged thoracic kyphosis and global positive imbalance were observed in D type compared to C type.

Conclusion C2–C7 A exhibited correlations with cervical balance and also with global balance. There should be various type of thoraco-lumbar alignment in DHS.

Graphic abstract

These slides can be retrieved under Electronic Supplementary Material.

Keywords Dropped head syndrome · Sagittal balance · Cervical deformity · Sagittal alignment

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Introduction

Dropped head syndrome (DHS) is characterized by chin-on-chest deformity which is caused by severe extensor muscle weakness and is passively correctable with neck extension

with normal passive neck range of motion [1]. This functional deformity arising from disability to balance the position of the head in standing or sitting posture impedes the quality of life of affected individuals significantly by inflicting various degrees of neck pain, difficulty in horizontal gaze, possible neurological symptoms, dysphagia or ambulation distress resulting in reduction in activities of daily living, and social interactions [2, 3]. There is a dispute about the sagittal alignment of DHS, which is essential for understanding the pathophysiology and planning treatment including operative corrective surgery. A previous study reported that cervical positive imbalance results in anterior translation of the center of gravity of the head and loads the weight of the head increasingly on neck extensors [2]. Therefore, sagittal malalignment of spinal elements may be related for the development of DHS. The primary aim of the present study was to clarify the characteristics in sagittal alignment of DHS through radiographic measurements.

Materials and methods

After obtaining institutional review board approval, a retrospective analysis of radiographic profiles was performed for the patients who presented to our department with chin-on-chest symptoms which were correctable in the supine position. All patients were referred to the Department of Neurology in our hospital to receive a neurological workup. Forty-one patients who were not found to have an underlying neurological condition and whose condition was diagnosed as isolated neck extensor myopathy (INEM) were included in this study: DHS may be confounded by a broad range of differential diagnosis such as Parkinson's disease [4], and INEM is considered to cause the chin-on-chest symptom in the absence of specific abnormality [5, 6]. All patients provided informed consent by opt-out manner. Lateral standing radiographs were obtained by using vertical film with a constant distance between the subject and the radiographic source with a radio-opaque calibration tool, following a standardized protocol with the patients standing in a neutral fists-on-clavicle position and instructed to look straight ahead with knees locked [7–12]. The following spinal parameters were assessed: (1) C2 slope (C2S) which was measured as the angulation of the intersection between lines parallel to the C2 inferior end plate and the horizontal line, (2) C2–C7 lordosis angle (C2–C7 A) which was measured as the angulation of the intersection between lines parallel to the inferior end plates of C2 and C7, (3) C2–C7 sagittal vertical axis (C2–C7 SVA) which was measured as the distance between the C2 plumb line and C7 supero-posterior corner, (4) T1 slope (T1S) which was measured as the angulation of the intersection between lines parallel to the T1 superior end plate and the horizontal line, (5) C7–S1 sagittal vertical axis

(C7–S1 SVA) which was measured as the distance between the C7 plumb line and S1 supero-posterior corner, (6) T1–T4 thoracic kyphosis angle (T1–T4 TK) which was measured as the angulation of the intersection between lines parallel to the T1 superior end plate and the T4 inferior end plate, (7) T4–T12 thoracic kyphosis angle (T4–T12 TK) which was measured as the angulation of the intersection between lines parallel to the T4 superior end plate and the T12 inferior end plate, (8) lumbar lordosis (LL) which was measured as the angulation of the intersection between lines parallel to the superior end plate of T12 and the sacrum, (9) sacral slope (SS) which was measured as the angulation of the intersection between lines parallel to the superior end plates of the sacrum and the horizontal line, (10) pelvic tilt (PT) which was measured as the angulation of the intersection between lines parallel to a line connecting the midpoint of the superior end plate of the sacrum to the center of the hip axis and the vertical line, and (11) pelvic incidence (PI) which was measured as the angulation of the intersection between lines parallel to a line connecting the midpoint of the superior end plate of the sacrum to the center of the hip axis and a perpendicular line to the superior end plate of the sacrum [13, 14]. The accuracy of the methods, including intra-rater and inter-rater agreements, was analyzed in detail in a previous paper [15]. We also measure chin-chest distance determined as the distance between the tip of jaw and the apex of the sternum.

Statistical analysis was performed using commercially available software (JMP version 11.0; SAS Institute, Inc., Cary, NC, USA). After determining that the data followed a parametric distribution using the Shapiro–Wilk normality test (where $P > 0.05$ suggests that the data are from a normal distribution), student t tests were calculated to compare the two groups (Diffuse kyphosis type: D type vs. Cervical kyphosis type: C type), and Pearson product-moment correlation coefficients were calculated for all combinations of radiographic measurements. The level of significance was set at $P < 0.05$. For significant correlations observed among radiographic parameters, further analyses, using both linear regression and logistic regression analyses with binary variables, were performed to determine a possible threshold of radiographic measurement was most significant.

Results

Patient characteristics

Twenty-seven female and 14 male patients with DHS with an average age of 73.0 ± 1.9 years (range, 42–87) who agreed to participate were enrolled in this study. The patients who had evidence of diffuse idiopathic skeletal hyperostosis inside or outside the cervical region were excluded from the analysis.

Radiographic measurements

Radiographic measurements are summarized in Table 1. C2–C7 SVA, which was representative of head balance, exhibited positive imbalance (52.0 ± 2.4 mm), determined by $C2-C7\ SVA > 40$ mm [16]. C2S and T1S were enlarged (C2S: $49.8^\circ \pm 1.8^\circ$, T1S: $33.1^\circ \pm 2.2^\circ$). Decreased C2–C7 A represents the kyphotic contour of DHS ($-8.7^\circ \pm 3.2^\circ$).

Statistical analysis

Correlations among radiographic measurements are summarized in Table 2. C2–C7 A exhibited significant correlations

Table 1 Radiographic measures of standing total spine

	DHS
C2S	$49.8^\circ \pm 1.8^\circ$
Chin-chest distance	59.1 ± 1.4 mm
C2–C7 SVA	52.0 ± 2.4 mm
C2–C7 A	$-8.7^\circ \pm 3.2^\circ$
T1S	$33.1^\circ \pm 2.2^\circ$
C7–S1 SVA	38.6 ± 9.0 mm
T1–T4 TK	$14.4^\circ \pm 2.1^\circ$
T4–T12 TK	$32.9^\circ \pm 2.9^\circ$
LL	$36.4^\circ \pm 2.9^\circ$
SS	$26.0^\circ \pm 1.8^\circ$
PT	$24.4^\circ \pm 2.0^\circ$
PI	$48.3^\circ \pm 2.3^\circ$

DHS, dropped head syndrome; C2S, C2 slope; C2–C7 SVA, C2–C7 sagittal vertical axis; C2–C7 A, C2–C7 lordosis angle; T1S, T1 slope; C7–S1 SVA, C7–S1 sagittal vertical axis; TK, thoracic kyphosis angle; LL, lumbar lordosis angle; SS, sacral slope; PT, pelvic tilt; PI, pelvic incidence

with thoraco-lumbar parameters (T1S: $r = 0.39$, $P = 0.01$; C7–S1 SVA: $r = 0.33$, $P = 0.04$; T4–T12 TK: $r = 0.39$, $P = 0.02$; SS: $r = -0.39$, $P = 0.02$, respectively). T1S exhibited significant positive correlations with cervical parameters (C2–C7 A: $r = 0.39$, $P = 0.01$; C2–C7 SVA: $r = 0.48$, $P = 0.002$, respectively) and thoraco-lumbar parameters (C7–S1 SVA: $r = 0.67$, $P < 0.001$; T1–T4 TK: $r = 0.33$, $P = 0.04$; T4–T12 TK: $r = 0.45$, $P = 0.004$; PT: $r = 0.36$, $P = 0.03$). C2–C7 SVA did not exhibit significant correlation with thoraco-lumbar parameter except for T1S.

For the strong correlation between C2–C7 A and C7–S1 SVA, both logistic and linear regression analyses were used to determine a possible radiographic threshold at which global positive imbalance occurs, which was defined by $C7-S1\ SVA > 50$ mm [17]. The logistic regression models predicted a value of -15.0 for C2–C7 A ($\chi^2 = 7.1$, $P = 0.008$) (Fig. 1) at which the P value for the correlation tests was lowest. Values of AUC calculated from ROC curves were estimated at 0.77 for the analysis, and seemed to accurately represent the results. The linear regression models predicted values of 6.0 for C2–C7 A ($r^2 = 0.11$, $P = 0.05$) (Fig. 2) responsible for a C7–S1 SVA value of 50 mm; C7–S1 SVA value of 50 mm represents global positive imbalance. Relative risk was also estimated at 2.9-fold (95% confidence interval [CI] = 1.0–8.8) in the case of $C2-C7\ A > -10$ which was a median value of C2–C7 A, and the odds ratio was estimated at 4.5-fold (95% confidence interval [CI] = 1.0–20.5) for the occurrence of global positive imbalance from the logistic analysis, where P value was estimated at 0.04.

Comparison between two types of DHS

For analysis obtained from the regression models, we classified DHS into two groups based on C2–C7 A, and set the

Table 2 Correlations among radiographic measures

	C2S	C2–C7A	C2–C7 SVA	T1S	C7–S1 SVA	T1–T4	T4–T12	LL	SS	PT	PI
C2S	1										
C2–C7A	−0.13	1									
C2–C7 SVA	0.11	−0.27	1								
T1S	−0.11	0.39*	0.48*	1							
C7–S1 SVA	−0.05	0.33*	0.24	0.67*	1						
T1–T4	0.12	0.05	0.20	0.33*	0.28	1					
T4–T12	−0.12	0.39*	0.12	0.45	0.62*	0.41*	1				
LL	−0.16	0.06	−0.22	−0.04	−0.23	−0.18	−0.09	1			
SS	0.02	−0.39*	0.05	−0.08	−0.10	−0.09	−0.06	0.68	1		
PT	−0.08	0.27	0.05	0.36*	0.44*	0.05	0.39*	−0.16	−0.33*	1	
PI	−0.06	−0.15	0.08	0.22	0.21	−0.02	0.26	0.48*	0.64*	0.51*	1

C2S, C2 slope; C2–C7 A, C2–C7 lordosis angle; C2–C7 SVA, C2–C7 sagittal vertical axis; T1S, T1 slope; C7–S1 SVA, C7–S1 sagittal vertical axis; LL, lumbar lordosis angle; SS, sacral slope; PT, pelvic tilt; PI, pelvic incidence

*indicates $P < 0.05$

Fig. 1 Bar graph of mean data

Fig. 2 Visual representation of the regression models superimposed on raw data. Positive correlation was observed between dropped head index (DH index) and C7–S1 sagittal vertical axis (C7–S1 SVA)

thresholds for C2–C7 A of -10° : the patients with C2–C7 A $> -10^\circ$ were classified into diffuse kyphosis type (D type) and others were classified into cervical kyphosis type (C type). Comparisons of radiographic measures between two groups are summarized in Table 3. These comparisons between two groups revealed the differences in thoracic contour. Kyphotic contour represented by enlarged T4–T12 TK was observed in D group (T4–T12 TK: $42.8^\circ \pm 3.1^\circ$ vs. $28.5^\circ \pm 3.3^\circ$).

Discussion

We observed enlarged offset of gravity of the head represented by C2–C7 SVA, and the correlation of C2–C7 A with C7–S1 SVA. For this correlation, we classified DHS into two groups based on C2–C7 A. The comparison analysis of two groups revealed there were two different type of thoracolumbar alignment in DHS.

Table 3 Comparison of radiographic measures between two groups

	D type	C type	P value
C2S	$49.4^\circ \pm 2.5^\circ$	$50.3^\circ \pm 2.7^\circ$	0.81
C2–C7 A	$3.2^\circ \pm 3.4^\circ$	$-22.6^\circ \pm 3.7^\circ$	<0.001
C2–C7 SVA	49.8 ± 3.2 mm	54.6 ± 3.5 mm	0.31
T1S	$37.9^\circ \pm 2.9^\circ$	$27.6^\circ \pm 3.1^\circ$	0.02
C7–S1 SVA	53.6 ± 11.3 mm	16.6 ± 11.3 mm	0.03
T1–T4 TK	$13.0^\circ \pm 2.6^\circ$	$13.0^\circ \pm 2.7^\circ$	0.23
T4–T12 TK	$42.8^\circ \pm 3.1^\circ$	$28.5^\circ \pm 3.3^\circ$	0.003
LL	$40.9^\circ \pm 3.3^\circ$	$34.4^\circ \pm 3.6^\circ$	0.19
SS	$25.3^\circ \pm 2.4^\circ$	$27.4^\circ \pm 2.6^\circ$	0.57
PT	$24.2^\circ \pm 2.2^\circ$	$21.6^\circ \pm 2.4^\circ$	0.43
PI	$49.1^\circ \pm 2.5^\circ$	$48.8^\circ \pm 2.7^\circ$	0.93

C2S, C2 slope, C2–C7 SVA, C2–C7 sagittal vertical axis, C2–C7 A, C2–C7 lordosis angle, T1S, T1 slope, C7–S1 SVA, C7–S1 sagittal vertical axis, TK, thoracic kyphosis angle, LL, lumbar lordosis angle, SS, sacral slope, PT, pelvic tilt, PI, pelvic incidence

It may be difficult to interpret these findings as actual causes of DHS because this was a cross-sectional study. However, some recent studies described the relationship between cervical sagittal alignment and other regions of the spinal column [18, 19]. With regard to a reciprocal relationship between the cervical and other parts of the spinal column, global sagittal imbalance leads to adaptive changes in the pelvis through a compensatory mechanism [18]. Hence, we assumed that the loss of head balance without global imbalance is a result of degenerative changes accompanied with anterior disk collapse at the middle and lower cervical spine, and named C type (Fig. 3). In contrast, the loss of balance with global positive imbalance is originated from global positive imbalance, which is named D type (Fig. 3). Finally in both types, with a progressing anterior shift of the head without balance and loss of tissue elasticity of the extensor muscle, the overstretched extensor muscles cannot maintain the head position in the standing or sitting position. Previously, sagittal spinal alignment in the DHS patient was simply described as a chin-on-chest deformity associated with cervicothoracic kyphosis. In fact, there should be various types of DHS alignment, as revealed in the current study.

Increased T4–T12 TK was observed in the D type compared to the C type. These results suggest the difference in affected muscles between the two types. The semispinalis capitis (originating from the T1–T6 transverse processes and C4–C6 articular processes, inserting at the occipital bone, innervated by posterior branches of spinal nerves), semispinalis cervicis (originating from the C7–T6 transverse processes, inserting at the C2–C5 spinal processes, innervated by posterior branches of spinal nerves), and semispinalis thoracis (originating from the T6–T12 transverse processes, inserting at the C6–T4 spinal processes, innervated by posterior branches of spinal nerves) are

Fig. 3 Representative standing radiographs of C type DHS. In C type, cervical imbalance is accompanied with severe cervical kyphosis and compensated by the spinopelvic complex, which shows balanced global balance. Patient: 73YO, Female. Sagittal parameters: C2S: 99°, Chin-chest distance: 36 mm, C2–C7 SVA: 73 mm, C2–C7 angle: -48° , T1S: 52° , C7–S1 SVA: -6 mm, T1–T4 TK: 29° , T4–T12 TK: 54° , LL: 42° , SS: 16° , PT: 30° , PI: 46°

C type

considered as the main extensors in the deep layer of the cervicothoracic region [20]. These semispinalis muscles may be responsible for the pathophysiology of INEM from the anatomical point of view. One report suggested the cause of DHS as the semispinalis capitis, in a middle-aged female patient with sepsis [21], which may tend to occur in the D type in light of the distribution of the muscle. On the other hand, the suboccipital group, which comprises

the rectus capitis posterior major, rectus capitis posterior minor, obliquus capitis superior, and obliquus capitis inferior (originating from C1–C2, inserting at the occipital bone, innervated by posterior primary divisions of spinal nerves), and spinalis cervicis (originating from the nuchal ligament, and the C7–T1 spinous processes, inserting at the C2 spinous process, innervated by anterior branches of spinal nerves) are a complex of the extensors in the deep

Fig. 4 Representative standing radiograph of D type shows the loss of head balance with relatively milder cervical kyphosis and shows diffuse positive imbalance. Patient: 78YO, Female. Sagittal parameters: C2S: 58° , Chin-Chest distance: 51 mm, C2–C7 SVA: 60 mm, C2–C7 angle: -5° , T1S: 56° , C7–S1 SVA: 86 mm, T1–T4 TK: 3° , T4–T12 TK: 52° , LL: 25° , SS: 23° , PT: 29° , PI: 52°

D type

layer of the cervical region and is considered affected in the C type for the distribution localized in the cervical region [22]. These differences might be considered as a target region in corrective surgery, and different strategies in conservative treatments are also required for each type. The cervical spine of the C type can be supported by only a cervical brace, but that of the D type requires an additional thoracic band in order to support the thoracic spine (Fig. 4).

The current study may have some limitations. First, the number of cases of INEM DHS is small, reflecting the small number of DHS patients in general, and the population may have skewed characteristics arising from inclusion criteria of this uncommon condition. Despite these limitations, we consider this study to provide some contribution to understanding the pathophysiology of DHS in view of performing optimal treatment.

Conclusion

Increased offset of the gravity axis of the head, which was represented by C2–C7 SVA, was observed in DHS. Among cervical parameters, C2–C7 A exhibited strong correlation with global balance. For these correlations, we made regression analyses on these parameters and classified DHS into two types according to the C2–C7 A value of -10.0 . The spinal sagittal alignment of D type shows thoracic kyphosis compared to C type.

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Compliance with ethical standards

Conflict of interest The authors declare no conflict of interest in any form received from a commercial party related directly or indirectly to this study.

Ethical approval This study was approved by the Ethics Review Committee of Tokyo Medical University Hospital. Written informed consent was obtained from all patients before the recruitment of patients in this study.

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