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Vertico-horizontal atlantoaxial index (VHAI): A new craniovertebral radiographic index

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ABSTRACT

Objective: The purpose of this study was to develop a new index that can reliably quantify the reduction of basilar invagination with atlantoaxial dislocation.

Patients and methods: Between May 2012 and September 2017, 40 patients with congenital basilar invagination and atlantoaxial dislocation as well as 100 sex-and age-matched control subjects were recruited for this study. All patients underwent direct posterior reduction and fixation. Mid-sagittal computerized tomography scan films were obtained before and after surgery as well as the vertico-horizontal atlantoaxial index (VHAI) was measured in all patients -before and after surgery- and controls. Additionally, the pre-and postoperative Japanese Orthopedic Association (JOA) scores, Nurick grading, European Myelopathy Score (EMS) and Prolo Scale score were used to evaluate the cervical myelopathy.

Results: The mean follow-up was 24.75 months with a range of 6–60 months. The mean value of VHAI in the control group was 87.86 ± 24.98 mm^2, while the mean values of VHAI before and after surgery were 209.45 ± 96.80 mm^2 and 95.08 ± 66.95 mm^2, respectively. Additionally, in the patient group, a negative correlation was observed between JOA, EMS, Prolo Scale scores and VHAI. On the other hand, a positive correlation was found between the Nurick grading and VHAI.

Conclusion: The VHAI can be an excellent measurement tool to evaluate the reduction of basilar invagination with atlantoaxial dislocation. There was a negative correlation between VHAI and JOA, EMS and Prolo Scale scores, and a positive correlation with Nurick grading; which indicates the effectiveness of this index.

1. Introduction

Basilar invagination is defined as a prolapse of the cervical spine into the cranial base and it is represented by the odontoid process invaginating into the foramen magnum [1–8]. It is often associated with other osseous anomalies of the craniovertebral junction, including an incomplete ring of C1, atlanto-occipital fusion, and hypoplasia of the atlas, basiocciput and occipital condyles [2,5,6,8–12]. Also, it is associated with neural axis abnormalities, including, syringomyelia, syringobulbia, hydrocephalus and Chiari malformation [2,4,5,8,12].

Basilar invagination is divided into two groups based on whether it is associated with atlantoaxial dislocation: 1. A group without atlantoaxial dislocation [1,4,8]. 2. A group with atlantoaxial dislocation (in which the odontoid process is dislocated posterosuperiorly and the foramen magnum becomes relatively narrow resulting in compression and injury of the medulla oblongata and/or spinal cord) [1,3,4,6–9,11–15]. Surgical management of basilar invagination with atlantoaxial dislocation focus on cervical spinal cord decompression, reduction and reconstruction of the atlantoaxial region [1,3,5,7,7,8,9,11,14–18]. In addition, there are many radiographic indices evaluating the subluxation, invagination and reduction of basilar invagination with atlantoaxial dislocation such as Chamberlain's line, McRae's line, Atlanto-dental Interval, etc.

In this study, we reported a new concept and technique -vertico-horizontal atlantoaxial index- to precisely quantify the relation of atlas and axis as well as accurately evaluate the reduction in patients with

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Table 1
Summary of clinical characteristics as well as pre- and postoperative values of VHAI, JOA, Nurick Grading, EMS and Prolo scale in 40 patients with Basilar Invagination and Atlantoaxial Dislocation.

Case No.	Sex/Age	Symptoms and signs	Duration of symptoms (months)	VHAI (mm^2)		JOA		Nurick Grading		EMS		Prolo Scale		Postoperative complications	Follow-up (months)
				Preop	Postop	Preop	Postop	Preop	Postop	Preop	Postop	Preop	Postop		
1	M/51	Neck pain; Paresthesia; Quadriparesis	0.5	237.14	42.38	12.5	14	3	2	13	16	6	8	None	24
2	M/61	Paresthesia; Urinary incontinence	156	230.17	87.77	10	13	3	2	13	15	6	8	None	16
3	M/52	Muscle weakness; Paresthesia	24	359.12	158.87	11	15	3	2	15	16	7	8	None	18
4	M/72	Dysphagia; Paresthesia	120	120.67	91.16	12	13	3	3	11	14	5	8	None	18
5	M/55	Occipitocervical Pain; Paresthesia; Muscle weakness	240	185.21	68.31	15	17	2	0	17	18	7	10	None	17
6	M/64	Paresthesia; Muscle weakness; Occipitocervical Pain	84	441.01	262.19	13	14	2	2	14	15	6	8	None	17
7	M/51	Neck pain; Paresthesia	24	202.73	93.79	15	17	2	1	15	18	8	10	CSF leakage	21
8	M/45	Neck pain; Paresthesia; Dysphagia	12	211.02	97.27	13	16	3	1	14	17	6	9	None	21
9	M/59	Paresthesia; Quadriparesis	2	337.02	218.15	12	13	3	3	14	15	6	7	None	16
10	M/61	Headache; Paresthesia;	12	203.25	60.85	14.5	15	3	2	13	14	6	8	None	16
11	M/26	Occipitocervical Pain; Muscle weakness; Dysbasia	24	370.08	95.99	14	16	3	1	14	15	7	8	None	26
12	M/44	Paresthesia; Dysbasia	48	97.56	80.27	15	16	3	1	14	15	7	9	None	26
13	F/18	Neck pain; Torticollis; Paresthesia	96	164.33	27.23	16	17	2	0	17	18	8	10	Wound infection	25
14	F/52	Paresthesia; Muscle weakness; Dysbasia	0.7	225.89	68.67	12	15	3	1	15	17	6	7	None	22
15	F/46	Neck pain; Paresthesia; Dysbasia	48	150.08	67.65	11	15.5	2	1	14	16	7	8	None	34
16	F/43	Headache; Dysesthesia	12	136.61	37.65	15	17	1	0	17	18	10	10	None	32
17	F/44	Dysesthesia; Occipitocervical pain; Muscle weakness	24	94.62	69.02	14	17	3	1	16	18	7	9	None	34
18	F/56	Dysesthesia; Dysphagia; Muscle weakness	24	257.84	157.26	13	15	3	1	14	16	6	8	None	35
19	F/46	Headache; Neck pain; Dysesthesia; Muscle weakness	24	143.38	78.46	14.5	16.5	3	1	14	16	7	10	None	36
20	F/43	Paresthesia; Occipitocervical pain	48	247.76	90.61	15	17	3	1	16	18	7	10	None	51
21	M/18	Paresthesia; Muscle weakness; Occipitocervical pain	24	395.32	226.68	10.5	14	3	2	13	15	7	8	None	46
22	F/42	Headache; Muscle weakness; Paresthesia	120	373.35	203.76	13	15	2	2	14	16	6	8	None	41
23	F/66	Paresthesia; Muscle weakness	18	326.43	301.82	13	14	3	2	14	15	6	7	None	57
24	F/58	Paresthesia; Dysbasia	120	252.44	26.75	14	16	3	1	16	17	7	8	None	54
25	F/57	Paresthesia; Occipitocervical pain	24	274.86	105.22	14	16.5	2	1	15	17	7	10	None	60
26	F/73	Muscle weakness; Dysesthesia; Occipitocervical pain	84	147.9	29.97	14	16	3	1	15	17	5	8	None	16
27	F/52	Occipitocervical pain; Transient loss of consciousness	24	129.04	50.76	16	17	3	0	16	18	7	9	None	16
28	F/65	Transient loss of consciousness; Dysesthesia; Quadriparesis	72	137.47	77.11	12	14.5	3	3	14	15	6	8	None	16
29	F/46	Muscle weakness; Dysbasia; Muscular dystrophy	2	309.15	140.93	13.5	17	1	0	16	18	8	10	None	7
30	M/43	Headache; Dysesthesia	24	133.66	58.46	14	17	1	0	15	18	8	10	None	18
31	M/52	Dysesthesia; Dysarthria	11	127.88	43.4	16	17	2	0	17	18	7	10	None	6
32	F/35	Muscle weakness; Dysesthesia; Muscular dystrophy	0.7	112.66	59.59	16.5	17	2	0	16	18	8	10	None	17
33	F/53	Muscle weakness; Paresthesia	3	184.06	94.72	15.5	17	2	1	14	18	6	8	None	20
34	F/40	Headache	60	142.03	77.39	17	17	0	0	17	18	10	10	None	21
35	F/40	Muscle weakness; Paresthesia	6	63.36	6.65	14	17	0	0	15	18	7	10	None	21
36	F/54	Dysbasia; Dysesthesia; Muscle weakness;	60	267.76	134.56	12	15	3	1	13	17	4	6	None	6
37	M/53	Muscle weakness; Paresthesia	24	136.3	45.08	14	17	1	0	15	18	8	10	None	10
38	F/44	Neck pain; Dysesthesia	104	140.67	80.5	16	17	1	0	16	18	9	10	None	22
39	F/50	Quadriparesis; Dysesthesia	4	249.33	51.93	12	17	2	0	14	18	7	10	None	22
40	F/50	Paresthesia; Headache	6	58.99	34.4	17	17	0	0	17	18	10	10	None	10

CSF: Cerebrospinal fluid, EMS: European myelopathy score, JOA: Japanese orthopedic association score, VHAI: Vertico-horizontal atlantoaxial index.

basilar invagination and atlantoaxial dislocation before and after surgery.

2. Patients and methods

2.1. Patient population

Between May 2012 and September 2017, 40 patients with congenital basilar invagination and atlantoaxial dislocation underwent direct posterior reduction and fixation technique that is described by Jian et al by using intraoperative reduction by distraction technique between Oc and C2 screws (no cervical traction was needed before or during the operation) [1]. The patient population comprised of 24 female patients and 16 male patients with ages ranging from 18 to 73 years (mean age: 49.20 years, standard deviation: 12.91 years). The odontoid process was displaced posteriorly compressing the upper cervical spinal cord and/or medulla in all patients. The clinical characteristics of all patients are summarized in Table 1.

Additionally, in order to obtain the normal range of vertico-horizontal atlantoaxial index (VHAI), mid-sagittal computed tomography (CT) scan films of 100 normal healthy control subjects, who underwent a negative CT workup after minor trauma, were analyzed as a control group. The control group comprised of 59 females and 41 males with ages ranging from 18 to 75 years (mean age: 51.57 years, standard deviation: 14.06 years) and showed no craniovertebral junction anomalies. In addition, the control subjects were matched for sex and age.

Patients with traumatic atlantoaxial dislocations, tumors, previous cervical surgery, os odontoideum, inflammatory pathologies such as rheumatoid arthritis, or known genetic syndromes such as achondroplasia, Down or Marfan syndromes were excluded.

In this study, basilar invagination was defined as the tip of the odontoid process exceeding the Chamberlain's line (CL) by 5 mm and atlantoaxial dislocation was defined as the value of atlantodental interval (ADI) exceeding 3 mm in adults [2,13,19,20]. The study was approved by the ethics committees of The First Affiliated Hospital of Chongqing Medical University, Chongqing, China. Furthermore, we used the routine preoperative consent as this was a retrospective clinical study. In addition, the study was performed in accordance with the STROBE (Strengthening the Reporting of Observational Studies in Epidemiology) statement [21].

2.2. Radiological evaluation

Thin-slice CT scans with three-dimensional (3D) reconstruction views were obtained before and on the first day after the operation for each patient while wearing a neck brace (PG 174 PROGLIDE® Universal Cervical Collars, USA) in a neutral position. All scans were obtained on a 128-slice multidetector CT scanner (Siemens, Germany). The acquired images were reconstructed in 3 orthogonal planes on a dedicated workstation. Additionally, the mid-sagittal plane was reconstructed by realigning the positioning crosshairs on the axial images. Images were analyzed on a preset bone window setting of a width of 2000 Hounsfield units and a length of 800 Hounsfield units.

2.3. Vertico-horizontal Atlantoaxial Index (VHAI)

In order to calculate the vertico-horizontal atlantoaxial index, a mid-sagittal computed tomography (CT) scan plane should be obtained. First, a horizontal line is drawn through the lower endplate of the axis (Red line 1, Fig. 1A). A second line is drawn parallel to this and tangential to the lower border of the anterior arch of the atlas (Yellow line 2, Fig. 1B). Further, a third line is drawn parallel to these lines and tangential to the superior margin of the dens (Yellow line 3, Fig. 1C). Next, a fourth line is drawn perpendicular to these lines and tangential to the posterior border of the odontoid process (Blue line 4, Fig. 1D). Finally, a fifth line is drawn parallel to line 4 -perpendicular to the first, second and third lines- and tangential to the posterior border of the anterior arch of the atlas (Blue line 5, Fig. 1E).

The shortest distance between the second and third lines called vertical displacement (VD). In addition, the shortest distance between the fourth and fifth lines called horizontal displacement (HD). Vertico-horizontal atlantoaxial index = vertical displacement × horizontal displacement (Fig. 1F).

All the measurements were performed by one of the authors (J.J.X., five years of experience with brain imaging) and an experienced independent neuroradiologist (11 years of experience with brain imaging).

2.4. Statistical analysis

Statistical analysis was performed using SPSS 22.0 (Chicago, USA) for Windows. A paired samples *t*-test was applied for comparing pre- and postoperative VHAI. In addition, an independent samples *t*-test was used for comparing the values of VHAI before and after surgery with the control group. The correlation between preoperative Japanese Orthopedic Association (JOA) scores, Nurick grading, European Myelopathy Score (EMS), Prolo Scale score and preoperative VHAI as well as postoperative JOA scores, Nurick grading, EMS, Prolo Scale score and postoperative VHAI were determined using the Spearman correlation test. A p-value of less than 0.05 was considered statistically significant. All data analyses were performed by an independent expert statistician (15 years of experience in statistical analysis).

3. Results

The means of vertical and horizontal displacement in the control group were 9.91 ± 1.94 mm and 9.83 ± 1.32 mm, respectively. On the other hand, the means in the pre- and postoperative groups were 12.37 ± 4.58 mm and 7.98 ± 3.73 mm, respectively, for vertical displacement, and 16.52 ± 2.95 mm and 10.97 ± 3.39 mm, respectively, for horizontal displacement.

Intraobserver and interobserver agreement were assessed using the intraclass correlation (ICC) test. The results in control subjects demonstrated excellent intraobserver and interobserver correlation coefficient (ICC = 0.97). In addition, the results in all patients before and after surgery also showed excellent intraobserver and interobserver correlation coefficient (ICC = 0.97 and ICC = 0.96, respectively).

Furthermore, the mean of the VHAI in the control group was 87.86 ± 24.98 mm^2. While, the pre- and postoperative means of the VHAI were 209.45 ± 96.80 mm^2 and 95.08 ± 66.95 mm^2, respectively. In addition, the pre- and postoperative means of Japanese orthopedic association score were 13.79 ± 1.78 and 15.85 ± 1.33, respectively. Moreover, the pre- and postoperative means of Nurick grading were 2.25 ± 0.95 and 1.00 ± 0.93, respectively. Additionally, the pre- and postoperative means of EMS were 14.80 ± 1.42 and 16.75 ± 1.35, respectively. Furthermore, the pre- and postoperative means of Prolo Scale score were 6.95 ± 1.30 and 8.83 ± 1.15, respectively. The pre- and postoperative VHAI, JOA, Nurick grading, EMS and Prolo Scale values are stated in Table 1.

The vertico-horizontal atlantoaxial index before surgery was significantly higher compared with the control group ($T = 4.76$, $p < 0.001$). On the other hand, there was no statistically significant difference between the VHAI values of the postoperative and control groups ($T = -0.65$, $p = 0.524$). Additionally, the vertico-horizontal atlantoaxial index was lower after surgery compared with the preoperative values ($T = 11.83$, $p < 0.001$). In addition, there was a negative correlation between the preoperative JOA score and preoperative VHAI ($r = -0.542$, $p < 0.001$) as well as the postoperative JOA score and postoperative VHAI ($r = -0.497$, $p = 0.001$). Furthermore, there was a positive correlation between the preoperative Nurick grading and preoperative VHAI ($r = 0.323$, $p = 0.042$) as well

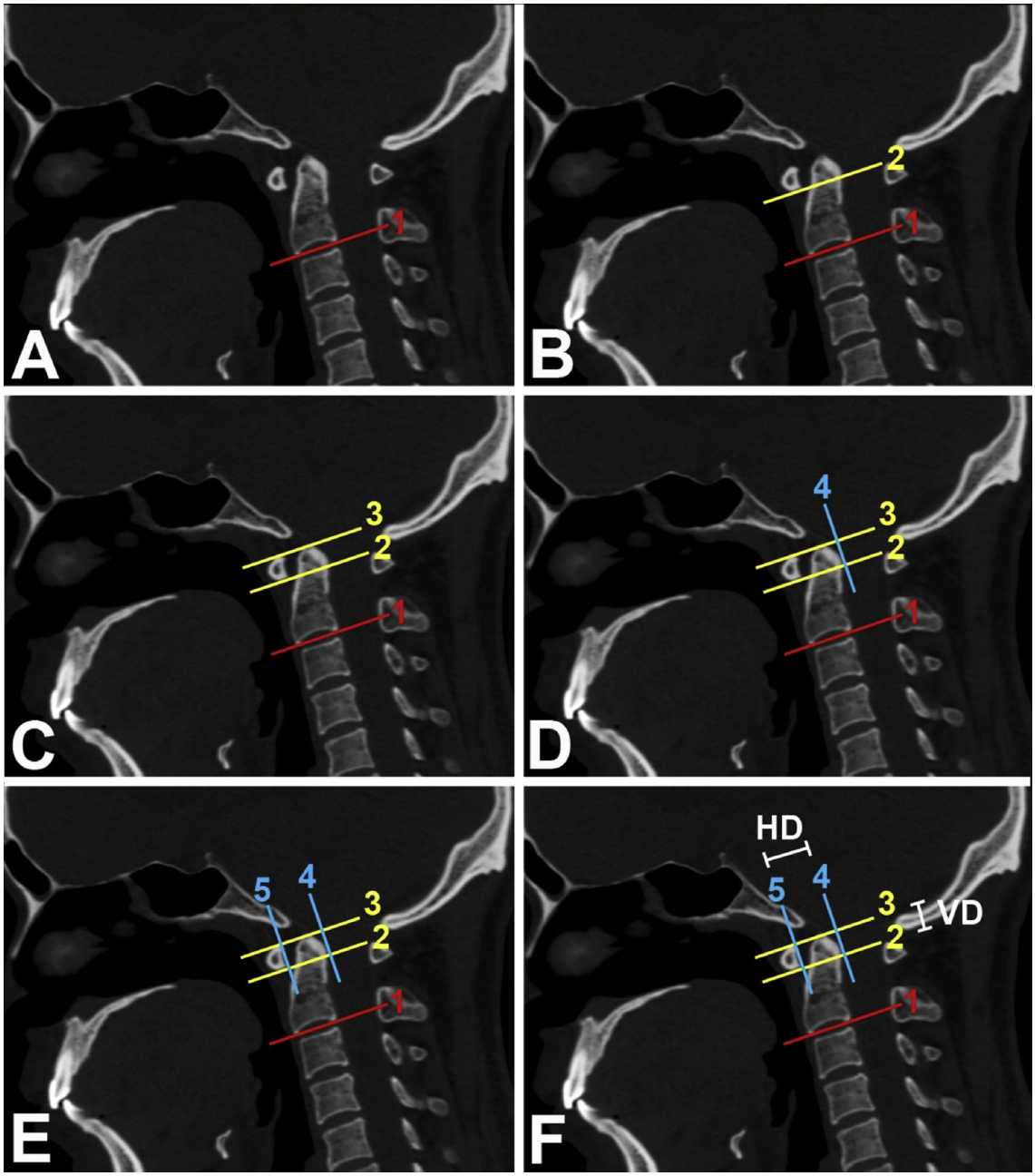

Fig. 1. Preoperative sagittal computed tomography (CT) scans demonstrating the required lines to measure the vertico-horizontal atlantoaxial index (VHAI) in a case of congenital basilar invagination with atlantoaxial dislocation. VD: vertical displacement, HD: horizontal displacement.

as the postoperative Nurick grading and postoperative VHAI ($r = 0.562$, $p < 0.001$).

As for European Myelopathy Score (EMS), there was a negative correlation between the preoperative EMS and preoperative VHAI ($r = -0.399$, $p = 0.011$) as well as the postoperative EMS and postoperative VHAI ($r = -0.483$, $p = 0.002$). Additionally, there was a negative correlation between the preoperative Prolo Scale score and preoperative VHAI ($r = -0.373$, $p = 0.018$) as well as the postoperative Prolo Scale score and postoperative VHAI ($r = -0.428$, $p = 0.006$).

All patients were followed-up for at least six months after surgery (6–60 months, an average of 24.78 ± 13.71 months) by the same neurosurgeon (J.J.X). In addition, there was a significant improvement of all clinical symptoms in all patients (100%) at the last follow-up (Table 1).

4. Discussion

In patients with basilar invagination and atlantoaxial dislocation, the point of maximal neural compression is the odontoid process. [3,6–8,22] Based on this pathological mechanism, neural compression can be divided into vertical displacement (along with the long axis of the odontoid process) and horizontal displacement (parallel to the lower endplate of the axis). Wherefore, the evaluation of neural compression can be achieved by measuring the displacement of the odontoid process in the mid-sagittal plane of CT images.

Several atlantoaxial distraction techniques have been reported and widely used, which may provide reduction and immediate stability simultaneously via a single posterior approach [1,4–7,10–12,15,18,23–26]. The single posterior approach preserves the anterior arch of the atlas and the odontoid process. Therefore, the vertico-horizontal atlantoaxial index is based and constructed on this

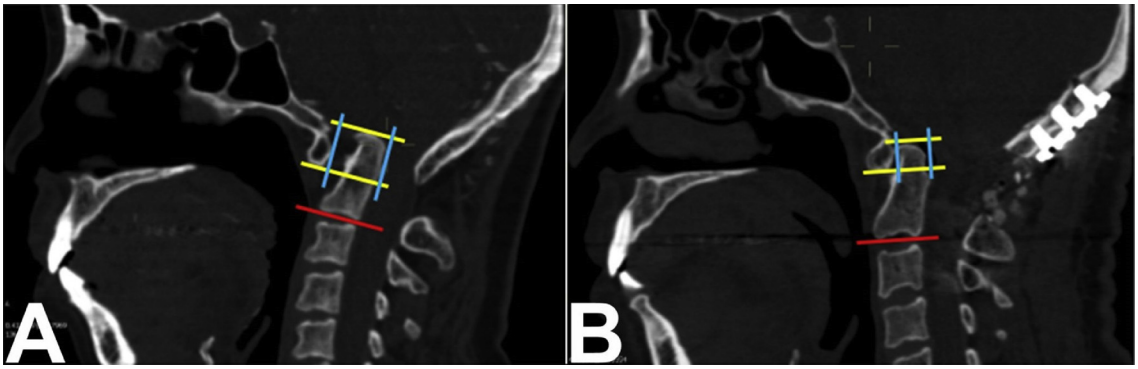

Fig. 2. Images obtained in a 57-year-old female patient who had two years of history of paresthesia and occipitocervical pain. (A): Preoperative sagittal reconstructed computed tomography (CT) scan showing the existence of basilar invagination with atlantoaxial dislocation and partial assimilation of the atlas (The VHAI was 274.86 mm^2). (B): Postoperative sagittal reconstruction CT scan showing a reduction of the dislocation (The VHAI was 105.22 mm^2). The reduction was performed using the direct posterior reduction and fixation technique. Duraplasty was not performed. Bone bridge formation is demonstrated between the occiput and the C2 spinous process.

approach. In this study, no patient needed a transoral decompression as a first-stage or second-stage operation.

The VHAI has a number of implications. Firstly, unlike all other craniovertebral measurements, VHAI is not an absolute number but a ratio. Hence, errors due to magnification are eliminated and it can be readily applied to radiographs and CT images. Secondly, it is primarily based on mid-sagittal CT images and hence errors due to superimposition of structures as in plain radiographs can be avoided (Fig. 2).

Patients with atlantoaxial dislocation often have different bony malformations like basilar invagination, atlanto-occipital fusion, atlas hypoplasia, Klippel-Feil syndrome, etc [6,8,9,11,13,14,20,27–30]. In the above-mentioned cases, evaluating the neural compression is difficult. There are many measurements for the evaluation of basilar invagination with atlantoaxial dislocation, reflecting the fact that no single method is consistent [17].

Chamberlain's line, McRae's line, McGregor's line, Fischgold's line, Redlund-Johnell method and Klaus height index are often used to observe the location of the odontoid process and the lower endplate of the axis. However, these methods are often interfered by the bony malformations like atlanto-occipital fusion. Furthermore, in Ranawat and modified Ranawat method, there are difficulties to precisely locate the midpoint of the anterior and the posterior arch of the atlas. While, with hypoplasia of the posterior arch of the atlas, the Sakaguchi-Kauppi method is difficult to measure. Further, these methods are based on plain radiographs; hence precise localization of landmarks may not be possible. Also, these methods cannot be measured postoperatively especially after resecting the posterior margin of the foramen magnum.

Additionally, the atlanto-dental interval can clarify the extent of horizontal dislocation of occipitocervical and atlantoaxial joints, however, it cannot correctly evaluate the backward extent of the odontoid process with a posterior tilt of the axis. Moreover, the basion-dens interval can display the backward extent of the dens, but it cannot correctly evaluate the vertical atlantoaxial dislocation [31]. Arvind et al. used the Vertical Atlantoaxial Index to quantify the degree of decompression which is based on the vertical subluxation of the axis in relation to the atlas, however; this technique cannot correctly evaluate the horizontal subluxation of the axis in relation to the atlas [17]. In addition, the cervicomedullary angle has been effectively used to assess the amount of basilar invagination, brainstem compression and the amount of postoperative decompression [32]. However, in patients with platybasia, there will often be no obvious changes in cervicomedullary angle after surgery [22,27].

In this study, all patients underwent a CT scan while wearing a neck brace (PG 174 PROGLIDE® Universal Cervical Collars, USA) in a neutral position. Indeed, the vertico-horizontal atlantoaxial index can be influenced by the neck positioning in patients with Type 2 atlantoaxial facetal dislocation based on Goel's classification of atlantoaxial facetal dislocation [31]. For that type of patients, the vertico-horizontal atlantoaxial index will be larger and smaller in flexional and extensional positions, respectively. Thus, in order to obtain the correct values of VHAI, all patients need to undergo CT scan in flexion and extension positions before and after surgery.

Computed tomography (CT) scan is preferable to observe bony changes compared to X-ray and magnetic resonance imaging (MRI) [33]. Furthermore, most of the previous measurements and methods have been performed based on plain X-rays. As for our index, it is based on CT images (Fig. 2). In addition, the VHAI does not depend on the posterior bony landmarks, such as the opisthion, which would be affected after concurrent suboccipital craniectomy for Chiari decompression (it offers a significant advantage over other radiographic measurements and methods of basilar invagination such as McRae's, McGregor's and Chamberlain's lines). Another problem with the aforementioned measurements and methods is that there is a wide variation in the normal values in healthy individuals.

As for evaluating the cervical myelopathy, several scores exist (the EMS, JOA, Cooper myelopathy scale, Prolo score and Nurick grading) that use various criteria to assess the grade of severity and the outcome after therapeutic intervention [34,35].

In 2008, Dalitz et al published a study that showed all 5 scores (the EMS, JOA, Cooper myelopathy scale, Prolo score and Nurick grading) are suitable for qualitatively evaluating the progression of cervical myelopathy and assessing the clinical characteristics [34]. Additionally, Vitzthum et al performed a retrospective study on 43 patients with cervical myelopathy and compared the outcome assessed by the 5 scores (the EMS, JOA, Cooper myelopathy scale, Prolo score and Nurick grading) [35]. They concluded that all 5 scores are internationally well established and showed a statistically significant correlation as well as measured postoperative improvement [35]. Furthermore, all the scores reflected clinical deficits of cervical myelopathy except for the Prolo score which rates the severity on the state of the economic situation above clinical symptoms [35]. Finally, they recommended using the Cooper myelopathy scale and EMS to evaluate the severity of the cervical myelopathy [35]. In addition, the Prolo score can be considered if the interest is focused on the regained ability to work and perform leisure time which may interest the paying authorities and public health [35].

On the other hand, Revanappa et al analyzed in their study the correlation between the Nurick grading and modified Japanese Orthopaedic Association (mJOA) in the pre- and postoperative follow-up in patients with cervical myelopathy. [36] The authors found a discrepancy between the Nurick grading and mJOA at follow-up evaluation and they explained it through the ability of patients to regain

employment without an improvement in the mJOA score [36]. Also, they recommended that both Nurick grading and mJOA score should be used in the evaluation of patients cervical myelopathy [36].

In our study, we used the EMS, JOA, Prolo score and Nurick grading.

Nevertheless, there are some limitations to the VHAI. First of all, it cannot be used in patients with os odontoideum and after surgery involving odontoidectomy. In addition, it's unable to address O-C1 distance which makes it less useful in patients where atlanto-occipital dislocation is a significant portion of the pathology. Also, we did not test it in children and cases with basilar invagination and atlantoaxial dislocation secondary to rheumatoid arthritis. Further, we only measured VHAI on CT scan because the bony landmarks can be determined more precisely compared to MRI. However, in the future, we will perform further studies to measure VHAI on MRI.

5. Conclusion

The vertico-horizontal atlantoaxial index is a reliable measurement tool to evaluate the reduction of basilar invagination with atlantoaxial dislocation. Also, it can estimate the relationship between atlas and axis. The VHAI showed a negative correlation with the JOA score, EMS and Prolo Scale score, and a positive correlation with Nurick grading; which indicates its effectiveness.

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