Inclination of the odontoid process in the pediatric Chiari I malformation

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Object. The quantitative analysis of odontoid process angulation has had scant attention in the Chiari I malformation population. In this study the authors sought to elucidate the correlation between posterior angulation of the odontoid process and patients with Chiari I malformation.

Methods. Magnetic resonance images of the craniocervical junction obtained in 100 children with Chiari I malformation and in 50 children with normal intracranial anatomy (controls) were analyzed. Specific attention was focused on measuring the degree of angulation of the odontoid process and assigning a score to the various degrees. Postoperative outcome following posterior cranial fossa decompression was then correlated to grades of angulation. Other measurements included midsagittal lengths of the foramen magnum and basiocciput, the authors’ institutions’ previously documented pB–C2 line (a line drawn perpendicular to one drawn between the basion and the posterior aspect of the C-2 body), level of the obex from a midpoint of the McRae line, and the extent of tonsillar herniation.

Higher grades of odontoid angulation (retroflexion) were found to be more frequently associated with syringomyelia and particularly holocord syringes. Higher grades of angulation were more common in female patients and were often found to have obices that were caudally displaced greater than three standard deviations below normal.

Conclusions. These results not only confirm prior reports of an increased incidence of a retroflexed odontoid process in Chiari I malformation but quantitatively define grades of inclination. Grades of angulation were not found to correlate with postoperative outcome. It is the authors’ hopes that these data add to our current limited understanding of the mechanisms involved in hindbrain herniation.

KEY WORDS • Chiari malformation • craniocervical junction • atlantoaxial joint • axis odontoid process • dens

A POSTERIORLY inclined odontoid process (odontoid process of epiphyses) of the axis has been noted in several clinicopathological conditions including the Chiari I malformation. The significance and extent of this anatomy, however, have not been thoroughly explored in patients with hindbrain herniation. Some authors have described a posteriorly tilted odontoid process as a normal variation that, on neuroimaging studies, can mimic a fracture. Our institution has previously defined parameters for anterior compression and its significance in Chiari I malformation. Investigators of this study, however, analyzed the relationship of the odontoid process to the basion and did not specifically measure the degree of posterior reclination that this process has nor its significance to outcome after posterior cranial fossa decompression. In the current study we retrospectively analyzed the degree of angulation of the odontoid process and its clinical sequelae both with regard to symptoms and outcome. Our hope is that these data will aid in the treatment of patients with Chiari I malformation and further elucidate evidence as to its pathophysiology.

Clinical Material and Methods

We retrospectively evaluated preoperative midsagittal T1-weighted MR images of the CCJ obtained in 100 patients aged 17 months to 18 years (mean age 9 years) in whom decompression of the Chiari I malformation was then performed by the senior author (W.J.O.) between 1992 and 2002. Forty-three patients were male and 57 were female. No patient had a history of significant cervical trauma. Syringomyelia was present in 52 patients. All of these patients underwent operative intervention; therefore, the natural history of the syrinx and odontoid tilt could not be determined. This evaluation focused on the angulation of the axial odontoid process, although the following measurements of the CCJ were also made: the length of the McRae line (see “a” in Fig. 1 left), distance of the obex to a midpoint of the McRae line (see “b” in Fig. 1 left), length of the odontoid process defined as a line drawn from the synchondrosis between the base and apical segments of the odontoid process to the apex of this same process (see “c” Abbreviations used in this paper: ANOVA = analysis of variance; CCJ = craniocervical junction; MR = magnetic resonance.
in Fig. 1 left), the midsagittal distance of cerebellar tonsillar tissue below the McRae line (see “d” in Fig. 1 left), a pB–C2 line (a line drawn perpendicular to one drawn between the basion and the posterior aspect of the C-2; see the article by Grabb, et al.,6 for details) (Fig. 1 center), the midsagittal distance of the basiocciput defined as a midpoint of the sphenoidoccpital synchondrosis to the basion (see “e” in Fig. 1 center), and the distance of the apex of the odontoid process from the line of Wackenheim (see “f” in Fig. 1 right). Two odontoid angles were measured. The first angle (Degree 1) was measured by drawing one horizontal line (to the plane of the image) through a midpoint of the synchondrosis between the base and apex of the odontoid process and the second line through the apex of the odontoid (see Angle “a” in Fig. 2 left). The second odontoid angle (Degree 2) was measured by drawing a horizontal line along the base of the axial body and another line through a midpoint of the apex of the odontoid process (Angle “b” in Fig. 2 right). Angulation based on our first odontoid angle technique (Degree 1), which reflected retroflexion of the odontoid process compared with retroversion, was next graded using the following scale: greater than 90˚, Grade 0; 85 to 89˚, Grade I; 80 to 84˚, Grade II (Fig. 3 left illustrates a case of Grade II angulation); and less than 80˚, Grade III (Fig. 3 right demonstrates a case of Grade III angulation). Measurements were performed using calipers and a goniometer. Each patient’s chart was then reviewed in an attempt to correlate clinical presentation and postoperative outcome with neuroimaging-defined anatomy. The two most common clinical presentations were headache and scoliosis. The mean follow-up period was 4.24 years (range 1 month–9 years). To serve as control data, these same measurements were made in 50 (23 male and 27 female) patients age 3 to 18 years (mean age 11 years) who had undergone T1-weighted MR imaging of the CCJ for a variety of reasons and in whom normal imaging results were demonstrated according to the radiologist’s report. Statistical analysis was performed using t-tests for comparison of means of distances and angles, and ANOVA was performed for nonparametric interpretation (p < 0.05, paired t-test, ANOVA, with Bonferroni correction for multiple comparisons). When appropriate, correlation coefficients were calculated. One observer (R.S.T.) made all measurements to avoid interobserver error, which can increase the coefficient of error to greater than 5%.

Results

In patients with the Chiari I malformation (defined as tonsillar ectopia of > 3 mm), a Grade 0 odontoid angle was documented in 16, Grade I in 30, Grade II in 22 (Fig. 3 left), and Grade III in 32 (Fig. 3 right). Of these angles, grades of II and III were observed in 57% of female and in 43% of the male patients. This skew toward female patients with higher angulation grades was statistically significant (binomial distribution analysis, p = 0.03). Four patients had basilar invagination. There were no os terminale (Bergmann tubercle), os odontoideum, evidence of odontoid fracture, odontoid hypoplasia, or dens bicorns identified. In one patient dolichoodontoid was found.23 Our second method (Degree 2) of measuring the angle of the odontoid (Angle b, Fig. 2 right) revealed a range of 52 to 87˚ (mean 70˚). The following are the ranges and means (in parentheses) in millimeters for pB–C2 distances for each odontoid angulation grade: Grade 0, -2 to 2.5 (1.2); Grade I, -1 to 5 (3.5); Grade II, 3 to 8 (5.5); and Grade III, 3 to 11 (6.8) (p < 0.01 when comparison was made between each grade and pB–C2). The mean lengths in millimeters of the odontoid process for each grade were as follows: Grade 0, 11.8; Grade I, 16; Grade II, 16; and Grade III, 17.2. Statistical significance of p < 0.05 was only seen when comparing Grades 0 and I. No significance was found when comparing odontoid lengths between sexes. The midsagittal distance in millimeters of the foramen magnum for odontoid angle Grades 0 through III had means of 29, 36, 36, and 36.5, respectively (p < 0.05 only between Grades 0 and
I–III). The standard deviation for each group was 2.1, 4.7, 3.8, and 4.1, respectively. Midsagittal distances in millimeters of the basiocciput for Grades 0 to III had means of 21, 24, 22, and 22, respectively. In nine patients an appreciated atlantoooccipital assimilation was present, in two of whom a Grade II angle was documented and in seven of whom a Grade III angle was determined. The Klippel–Feil anomaly was seen in four patients, two with Grade II angulation and two with Grade III angulation. Mean distances in millimeters from the inferior tip of the cerebellar tonsils to the McRae line for each grade were as follows: 9.6, 11.4, 11.1, and 14.4, respectively (p < 0.05 with comparison of Grades 0–II, and p = 0.001 for Grades II and III). There was no statistical significance (p > 0.05) in the morphology (pointed compared with rounded) of the cerebellar tonsils for each grade. Of all 100 patients a rounded tonsillar configuration was found in 15%. For those with Grades 0 and I odontoid angles syringomyelia was identified in six patients each. Of those with Grade II, 14 patients harbored a syrinx; of those with Grade III, 26 patients harbored a syrinx. Comparison of all odontoid angle grades and correlation with syringomyelia was statistically significant with p > 0.05. Correlation between grade of odontoid angulation and midsagittal length of the foramen magnum was only found if one compared Grades I through III odontoid angulations, which had greater mean distances than controls (r = 0.8).

In control individuals, odontoid angulation (as measured with Angle a, Fig. 2 left) ranged from −89° (Grade I) to +101° (Grade 0) (mean +97°). Grade 0 odontoid angles were found in 96% of controls. The remaining 4% were all Grade I odontoid angles. Angle b (Fig. 2 right) measurements ranged from 60 to 105° (mean 95°). The length of the odontoid process in controls ranged from 11 to 17.5 mm (mean 15.8). The range for midsagittal distances of the foramen magnum and basiocciput was 37.5 to 42 mm (mean 40 mm) and 17 to 27 mm (mean 23 mm), respectively. All obices were found to be greater than 2.5 mm superior to the plane of the foramen magnum (range 7–13 mm, mean 10 mm). In all control individuals there was no odontoid tip that violated the line of Wackenheim. Table 1 provides a summary of measurements obtained in all 100 individuals.

The most common clinical manifestations of Chiari I malformation in our group of 100 children were headache and scoliosis with syringomyelia. No correlation was found between either of these conditions or any other symptom and the degree of odontoid angulation—that is, there was no prevalence of any one symptom in any grade of odontoid angulation (ANOVA, p < 0.05). There were only three patients in whom syringomyelia persisted following posterior fossa decompression in our series. Of these, the odontoid process was angled posteriorly by 4, 10, and 14°. In each case the syrinx diminished in size after a second posterior fossa procedure in which unilateral tonsillar coagulation was performed.

No one grade of odontoid angulation correlated with the successful alleviation of preoperative symptoms—that is,
a patient with a Grade 0 angulation was just as likely to experience relief of preoperative symptoms as one with a Grade III.

Discussion

Ontogenetically there is metameric segmentation between the odontoid process and body of the axis as in the rest of the human vertebral column. Embryologically, the apex of the odontoid process is derived from the proatlas (fourth occipital sclerotome) and the primitive atlas (caudal portion of the fourth occipital and cranial portion of the first cervical sclerotome).27 Ossification proceeds from two lateral centers in the odontoid process, which become established during the 5th fetal month. These centers are separated by the notochord as it projects toward the basion via the apical ligament.22 At birth these centers are fused and ossification proceeds cranially. Furthermore, the apex of the odontoid process is often formed from a further center, the ossiculum terminale Bergmann, which can frequently assimilate, basilar invagination, and retroversion of the axial odontoid process. Some writers have speculated that the osseous anomaly in the Chiari I malformation is the initial deformation that is followed by the hindbrain herniation through the foramen magnum.19

Odontoid Process Angulation

Historically, many techniques have been conducted to measure the angle of the odontoid process. Both radiographic3,12,18 and dry-specimen2,35 methods have been used. With multiple techniques, however, come multiple results in ranges of normality. In fact, some investigators have discussed the necessity of some degree of reclination of the odontoid process for the normal biomechanics of flexion and extension of the atlantoaxial joint.23 In a study of 51 dried human C-2 vertebrae, Doherty and Heggeness2 found the odontoid process angle in the sagittal plane varied the most among all measurements. We have used two methods of measuring the angulation of the odontoid process in this study (Fig. 2). Our first method of angle measurement (Fig. 2 left) more accurately addressed retroflexion of the odontoid compared with retroversion as with the second method. Our second method (Fig. 2 right), however, was found to vary little and had the propensity to change with alterations of the C-2 disc or body base. We believe retroflexion of the odontoid to be more prevalent in the Chiari I malformation population and, as in this study, to be more likely to cause symptoms. Milhorat, et al.,16 found that in 26.4% of their patients with determined in the present study were based on midsagittal images obtained in children, they were measured from the neural central synchondrosis to the odontoid apex. We found no statistical difference in odontoid lengths when comparing sexes, controls, or odontoid angles.

Many hypotheses have been posed for the origin of the Chiari I malformation. In many of these patients a smaller than normal posterior fossa volume has been noted16,22,23. We could not analyze the posterior fossa volume in this retrospective study, but did find that the basiocciput was not statistically shorter in the midsagittal plane compared with controls for each age (p > 0.05). There were no statistical differences between grades of odontoid angulation and the length of the basiocciput. Lange23 has shown that the midsagittal distance of the basiocciput has a mean range of 13.1 to 19.1 mm in children age 0 to 6 years. Our mean range for this age group was 16 to 25 mm, which indicates that the basiocciput was not particularly underdeveloped in this group with hindbrain herniation. Presence of a small posterior fossa has been proposed as the predisposing factor of hindbrain herniation, although this cavity has been found to lie within normal limits in many patients with tonsillar ectopia.24,33 and, like Chiari II malformations, may be noted in patients with the Chiari I malformation.16 Other authors have discussed additional osseous anomalies in association with hindbrain herniation.15 These include atlantooccipital assimilation, basilar invagination, and retroversion of the axial odontoid process. Some writers have speculated that the osseous anomaly in the Chiari I malformation is the initial deformation that is followed by the hindbrain herniation through the foramen magnum.19
Odontoid process in pediatric Chiari malformation

TABLE 1
Summary of distances and angles stratified by angulation grade for patients with Chiari I malformation*

<table>
<thead>
<tr>
<th>Measurement</th>
<th>All Malformations (100)</th>
<th>0 (16 cases)</th>
<th>I (30 cases)</th>
<th>II (22 cases)</th>
<th>III (32 cases)</th>
<th>Controls (50 cases)</th>
</tr>
</thead>
<tbody>
<tr>
<td>distance/length (mm)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>a</td>
<td>35.1 ± 5.1</td>
<td>29 ± 2.1</td>
<td>36 ± 4.7</td>
<td>36 ± 3.8</td>
<td>36.5 ± 4.1</td>
<td>40 ± 5</td>
</tr>
<tr>
<td>b</td>
<td>2.1 ± 2.5</td>
<td>3.4 ± 3</td>
<td>2.7 ± 3.3</td>
<td>0.1 ± 2.5</td>
<td>−2.4 ± 3.7†</td>
<td>10 ± 6.2</td>
</tr>
<tr>
<td>c</td>
<td>15.8 ± 3.8</td>
<td>11.8 ± 3</td>
<td>16 ± 3.1</td>
<td>16 ± 2.5</td>
<td>17.2 ± 2.1</td>
<td>15.8 ± 4</td>
</tr>
<tr>
<td>d</td>
<td>12.6 ± 7</td>
<td>9.6 ± 3</td>
<td>12 ± 4.1</td>
<td>13 ± 5</td>
<td>14 ± 3.1</td>
<td>NA</td>
</tr>
<tr>
<td>e</td>
<td>23 ± 4.1</td>
<td>21 ± 5</td>
<td>24 ± 3.9</td>
<td>22 ± 3.4</td>
<td>22 ± 4</td>
<td>23 ± 6.9</td>
</tr>
<tr>
<td>Angle a (°)</td>
<td>98 ± 6</td>
<td>100 ± 5</td>
<td>86.6 ± 1.1</td>
<td>82 ± 1.3</td>
<td>77 ± 3.1</td>
<td>97 ± 5.1</td>
</tr>
<tr>
<td>Angle b (°)</td>
<td>70 ± 9.2</td>
<td>85 ± 6.3</td>
<td>74 ± 3.6</td>
<td>62 ± 4</td>
<td>55 ± 4.8</td>
<td>95 ± 6.6</td>
</tr>
</tbody>
</table>

* All letters and angles correspond to those depicted in Fig. 1 left and center. Abbreviation: NA = not applicable.
† In using Angle a, this value denotes an obex inferior (−) to the plane of the foramen magnum.

a Chiari I malformation an odontoid process that was retroflexed was present. This was based on the angulation (> 15°) of the odontoid compared with a line connecting the basion to a midpoint of the axial body, which, in our experience, has a configuration that can vary widely especially in the population with hindbrain herniation. In comparison, Goel, et al.,4 have used a modified omega angle for describing a retroverted odontoid in patients with basilar invagination and Chiari I malformation. This method also has the inherent flaw of depending on the irregular base of the axis. In an elaborate analysis of odontoid angulation, Krmpotić-Nemanić and Zagreb10 have demonstrated that the kyphosis of the skull base and the kyphosis of the odontoid process together compensate for the lordosis of the cervical spine. For example, with increased angulation of the skull base (sphenoidal angle) the odontoid process will become lordotic to compensate for this kyphotic deformity of the skull base. Koebke6 has shown that the straight odontoid process is only subjected to stress due to compression whereas the dorsally inclined odontoid process is subjected to bending stress that modifies its trabecular system with two curved bundles of trabeculae intersecting each other at right angles. This lordotic odontoid process configuration consequently has a transverse ligament that in cross-section is rather narrow. To date, angulation of the odontoid process has had no significant quantitation in the Chiari I malformation. Gosain, et al.,3 found that the posterior inclination of the odontoid process, with respect to the foramen magnum, appears to be the best indicator that a patient is at risk for basilar impression. In two of our patients we found an odontoid apex that violated the McRae line and in two others an odontoid apex that rose superior to the clival line of Wackenheim. Only one of these patients required a subsequent transoral odontoidectomy. Grabb, et al.,6 have reported that anterior compression at the cranio cervical junction does not correlate with the presence of syringomyelia in the Chiari I malformation and that this compression was seemingly due to a posteriorly oriented odontoid, not true basilar invagination. Our present results show that higher grades of odontoid angulation do indeed correlate with syringomyelia: a syrinx was apparent in 74% of patients with a Grade II or Grade III odontoid angulation. Of holocord syringes, 70% were demonstrated in patients with a Grade III odontoid angulation. Curiously, no single grade of odontoid angulation correlated with whether a patient would experience relief of preoperative symptom(s).

The odontoid process mechanically moves more posteriorly with cervical flexion, which with a posteriorly inclined bony process can exaggerate basilar coarctation because the cerebellar tonsils are more inferiorly located with flexion of the cervical spine.21 Maximum flexion and extension in the occipitaxial region, however, do not correspond to maximum flexion and extension of the neck. For example, maximum posterior tilt of the odontoid process is not seen with mere flexion of the cervical spine but rather with the head drawn posteriorly and the chin tucked inwardly.21 This would be maximized in our patients with a Grade III odontoid angulation. Interestingly, in female patients higher grades of odontoid angulation were more likely, but McRae and Barnum14 found that atlantooccipital assimilation was more common in males. In our series, assimilation was seen in four female and five male patients.

Compared with the findings of Grabb, et al.,6 our Grade II and III odontoid tilts were associated with mean pB–C2 measurements of 5 and 6.5 mm, respectively. It is intuitive that a more posteriorly inclined odontoid would tend to cause more deformation at the CCJ. We found, however, no specific symptom to correlate with each grade of reclamation. This could merely be due to the fact that our grade of odontoid angulation was purely an osseous measurement and did not account for additional accompanying soft-tissue compression. Patients with a pB–C2 distance of 9 mm or greater will probably require not only a posterior fossa decompression but also odontoidectomy.6 Our hypothesis would be that a Grade II or higher odontoid angulation does not necessarily equate with anterior brainstem compression in the face of a capacious CCJ.

Are patients with exaggerated cervical lordoses more prone to hindbrain herniation? Without clear standardized methods of measuring spinal curvature on lateral radiographs this is difficult to address. Four of our patients had known Klippel–Feil syndrome, which is more prone to an increased cervical lordosis24 and appears to have a higher incidence in the Chiari I malformation population.22 Could this be one reason for the higher incidence of this anomaly...
in the Chiari I malformation population, in that the reversal of the cervical curvature pushes the odontoid process posteriorly thereby predisposing these patients to hindbrain herniation? The incidence of the Chiari I malformation, however, is not known. Functionally, one could entertain that a posteriorly tilted odontoid process displaces the brainstem posteriorly at the level of the obex, which then “opens up” the foramen cecum and inhibits cerebrospinal fluid egress into the subarachnoid space, thus propelling it more directly into the spinal cord and increasing the incidence of syrinx formation (which is seen in 50–75% of this population). This mechanism for syrinx formation has been proposed by Gardner and Angel1 but not with specific regard to odontoid displacement. Yamazaki, et al., have reported that in patients with hindbrain herniation not associated with syringomyelia compression of the brainstem may be more pronounced at the CCJ. Whereas we also cannot conclude that posterior angulation of the odontoid process predicates hindbrain herniation in the Chiari I malformation, the fact that a posteriorly inclined process was found in 84 of our patients (30% with Grade III angulation) and in only 10% of controls suggests that this process is at least an amplifier if not a cause of a significant degree of tonsillar ectopia. The obices in this study were greater than three standard deviations below the normal position and were directly correlated to greater grades of odontoid angulation (p = 0.00047). If one considers greater amounts of soft tissue (tonsils, brainstem) and their mass effect, then instinctively the odontoid angulation would be more anteriorly displaced compared with our findings of more posteriorly displaced processes with greater amounts of soft tissue (that is, brainstem) at the CCJ. An inferiorly displaced obex has been seen in the various Chiari malformations.20,26,31 This displaced brainstem consequently adds to the compression at the CCJ in the Chiari I malformation.

Conclusions

Although measurements based on radiographs are subject to artifacts of magnification, rotation, and projection, they are often clinically useful. We have found that higher grades of posterior angulation (Grades II and III) as measured on midsagittal MR imaging are more often associated with syringomyelia and in particular holocord syringes. This was specifically with regard to retroflexion and not retroversion of the odontoid process (the former defined as posterior angulation of the odontoid process by measuring Angle a [Fig. 2 left] compared with retroversion defined as posterior angulation of the odontoid process by measuring Angle b [Fig. 2 right]). Higher grades were found more often in female patients. We have also shown that the obex is more caudally displaced in association with higher grades of odontoid angulation. Interestingly, no single grade of odontoid angulation correlated with resolution or partial relief of preoperative symptoms. These data add to our knowledge base that osseous anomalies of the CCJ are often observed in patients with Chiari I malformation. Whether these lesions predate, cause, or exaggerate the hindbrain herniation, however, remains to be elucidated.

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