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Original Article

Increase of the clivus-canal angle in patients with basilar invagination, without atlantoaxial displacement, treated with a simple maneuver of indirect decompression of the odontoid with the head clamp, during posterior occipitocervical arthrodesis

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ABSTRACT
Background: Basilar invagination (BI) can be defined as the insinuation of the content of the craniovertebral junction through the foramen magnum toward the posterior fossa. BI is a prevalent condition in Northeast Brazil. The present study describes the changes in the clivus-canal angle (CCA) in the postoperative period in patients with symptomatic BI operated by a posterior approach, using a simple technique of indirect reduction of the odontoid associated with occipitocervical fixation.

Methods: Patients underwent radiological evaluations by magnetic resonance imaging in the pre and postoperative periods, where the height of the odontoid tip was measured in relation to the Chamberlain line and the ACC. All patients underwent posterior occipitocervical fixation with specific maneuvers of distraction and extension of the cephalic segment with the aid of a head clamp with three fixation points for anterior reduction of the odontoid.

Results: Among the 8 patients evaluated in the series, all had increased ACC in the postoperative period, with a mean of 14.81 ± 1.54°, and statistically significant difference between the pre and postoperative periods (\(P < 0.05\)).

Conclusion: The indirect surgical reduction of the odontoid process by a posterior approach through the manipulation (distraction-extension) of a “Mayfield” type of head clamp followed by occipitocervical fixation proved to be effective in improving the ACC, being easily reproducible.

Keywords: Arnold Chiari malformation, Foramen magnum, Odontoid process, Skull base, Sphenoid bone

INTRODUCTION
Basilar invagination (BI) is part of a larger spectrum of malformations in the development of the craniovertebral junction (CJV) and can be defined as the insinuation of the content of the CJV through the foramen magnum toward the posterior fossa.[3,5,7,16,36] Classically, BI
has been described radiologically based on the height of the tip of the odontoid process in relation to the Chamberlain line (5 mm above).\[2\] Goel classified BI into two subtypes according to the presence of atlantoaxial displacement (AAD): Type A, where there is AAD and the odontoid tip is inserted through the magnum foramen and Type B, where AAD does not exist and the odontoid tip does not cross the lower border of the clivus.\[22\] BI is frequently found in the Northeast of Brazil, and in this location, it is associated with Chiari malformation (Types 1 and 2) in 80% of the cases and less frequently with syringomyelia, belonging to Goel Type B.\[9,10,11,39\]

Symptomatic BI is usually a progressively debilitating condition and its failure to be recognized and treated can predispose patients to develop disabling neurological deficits that can proceed until death.\[36\] Although it is a surgical condition, there is no agreement on the most appropriate technique or even the surgical route to be used (anterior versus posterior).\[3,12,22,30,31,38,40,41\] Apparently, there is no superiority between the different techniques.\[36\]

In the present study, the authors describe the changes in the clivus-canal angle (CCA) in patients with “Goel type B” BI, after the application of a simple surgical maneuver to reduce indirectly the odontoid peg, by a posterior approach alone, with the user of a Mayfield head clamp and a posterior occipitocervical fixation (POCF).

**MATERIALS AND METHODS**

**Patients**

The local review board approved the study protocol (CAAE-77722017.7.00005200) and all patients gave written consent for data analysis. The study was a retrospective review of medical records of patients with symptomatic BI, with surgical indication, and who underwent the technique under analysis between 2015 and 2017, in a single institution and with the same surgeon (CV).

The inclusion criteria were as follow: (a) primary BI; (b) symptoms and signs compatible with compression at the foramen magnum such as: ataxias, pyramidal, sensitive, and lower cranial nerve dysfunctions; and (c) tip of the odontoid process 5 mm above in relation to the Chamberlain’s line during magnetic resonance imaging (MRI) evaluation.

The following exclusion criteria were adopted: (a) patients not submitted to POCF; (b) patients previously submitted to odontoidecotomy or posterior fossa bone decompression; (c) injuries to CVJ secondary to trauma, neoplasia, or inflammatory diseases; and (d) unavailability of imaging exams in the pre or postoperative periods.

**Procedures**

**Surgical technique**

After anesthetic induction, the “Mayfield” head clamp (HeadFix Micromar, São Paulo) is installed with the three pins, which should be in the same axial plane at the level of the parietal eminences and upper temporal line. Afterward, the patient is placed in prone position. It is extremely important to ensure that the patient's trunk is fixed to the operating table with strips of adhesive tape [Figure 1a and b]. Fixing the patient to the table allows the cranial segment to be pulled in relation to the trunk. Initially, the head is positioned, above the heart, with slight flexion to facilitate CVJ bone decompression and placement of the subaxial screws of the lateral mass.

After skin incision and dissection through the midline avascular planes, the posterior bone elements of the occipital bone, CVJ, and subaxial spine are exposed. In this study, all patients were submitted to craniectomy of the occipital bone with the opening of the foramen magnum and removal of the posterior arch of C1. The dura mater of the CVJ was also opened and expanded with an autologous pericranium graft. No patient underwent aspiration or resection of the cerebellar tonsils.

Lateral mass screw was positioned bilaterally, including levels C3, C4, and C5, using the Roy-Camille technique. The deformity associated with BI in this series was so pronounced that the insertion of screws at levels C1 and C2 was not possible due to the impaction at the skull base of these segments and the large associated anatomical variations.

The surgeon positioned himself in front of the patient’s head, and the assistant loosened the screw locks on the head clamp’s rails and unlocked the Mayfield lever [Figure 1c-e]. The distraction-extension maneuvers of the craniovertebral segment were then applied to the cranial fixator [Figure 1f], allowing the displacement of the device through the tracks of the operating table. The position was considered appropriate when the patient’s line of sight was perpendicular to the floor [Figure 1g]. The transoperative radiological control was not adequate due to the superposition of the bone structures at the CVJ, which becomes more accentuated in patients with BI. After that, the Mayfield was locked in reverse order by the assistant.

A rod-plate POCF system was then inserted bilaterally, and after its manual molding in the CVJ region, the system was blocked with the use of locking caps in the subaxial region and bicortical screws in the occipital bone [Figure 1h]. Small fragments of autologous bone from the suboccipital craniectomy and from the C1 posterior elements were placed in the CVJ region, after decortication of the bone surfaces, laterally to the osteosynthesis material.
The procedure was completed by firstly closing the aponeurotic galea, then the subcutaneous and the skin, thereby reducing dead spaces and the formation of collections. No drain was used.

**Radiological evaluation**

All patients underwent an MRI in the pre and postoperative periods using a 1.5 Tesla scanner (Intera, Philips Medical Systems, Best, The Netherlands). The odontoid height (OH), in centimeters, was defined by the height of the odontoid tip in relation to the Chamberlain line. It was calculated in all patients only in the preoperative period, as the patients underwent suboccipital craniectomy. The clivus-canal angle (CCA) was measured, in degrees, using median sagittal slices in the T1 sequence, and compared in the pre and postoperative periods. The CCA was obtained by measuring the angle between drawn lines tangent to posterior surfaces of the clivus and odontoid process, respectively [Figure 2]. Both parameters were calculated using the program Horos™ image viewer version 3 (Horos Project, licensed under GNU Lesser General Public License, United States of America).

**Statistical analysis**

All data were stored on an Excel 2010 worksheet and statistical calculations were performed on PRISM®, version 6 for Windows (GraphPad Software Incorporated). The data were presented in terms of absolute and percentage values and measures of descriptive statistics such as: mean, standard deviation, minimum, and maximum values. For comparison between the pre and postoperative periods, the paired t-test was used. The margin of error adopted was 5.0%.
RESULTS

Initially, data from 22 patients with CVJ diseases operated on by one of the authors (CV) was collected at a single institution between March 2015 and July 2017. Of these, 14 patients were excluded from the study, 9 for not having undergone to POCF, and 5 for not having sufficient radiological data, leaving only 8 patients for analysis.

In relation to sex, five patients (62.5%) were women and three (37.5%) were men. The minimum age was 33 years and the maximum 68 years, with a mean of 49.25 ± 11.29 years. All patients in the study had associated Chiari type 1 malformation.

The mean of OH was 2.87 ± 0.3 cm. The minimum and maximum values were 2.5 and 3.3 cm, respectively. The CCA in the preoperative period varied between 66 and 100.7°, with a mean of 83.6 ± 13.1° [Table 1].

In all patients, an increase in CCA was observed in the postoperative period. The mean of this increase was 14.81 ± 1.54°, ranging from 4.9 to 40.8°. There was a statistically significant difference between the pre and postoperative periods in relation to CCA (P = 0.0073, paired t-test).

Surgical postoperative complications that required active interventions occurred in two (25%) patients. One patient developed cerebrospinal fluid leakage on the fifth postoperative day, requiring a new surgical intervention for dural graft suture. Another patient developed meningitis in the postoperative period and was treated with a broad-spectrum antibiotic regimen for 14 days. Both had no additional sequelae. Two patients still had transient horizontal nystagmus, observed only in the first 48 h of the postoperative period. There were no deaths.

DISCUSSION

BI is a prevalent condition in the Northeast of Brazil, associated with other craniometric changes such as platybasia and brachycephaly.[9-11,18,33,39] Chiari malformation is present in about 80% of these patients. With these characteristics, patients are classified as belonging to type B according to Goel's classification, and the deformity is considered irreducible based on the application of external traction.[21,22] Subtype A of Goel is the most prevalent in the Indian subcontinent and characterized by the occurrence of AAD and insinuation of the odontoid through the magnum foramen. This is quite unusual in the population where the study was carried out.[21]

Goel proposed that type B patients of his classification should be submitted to posterior decompression of the foramen magnum as a fundamental step in their treatment.[21] As the BI is understood as a vertical instability of the CVJ, the POCF is usually added, considering that nearly one-third of the patients submitted only to decompression may have an early or late clinical worsening.[28,43] Classically, symptomatic nonreducible BI has been treated by circumferential

| Patient | Age (years) | OH (cm) | Preop CCA (°) | Postop CCA (°) |
|---------|-------------|---------|---------------|---------------|
| 1       | 54          | 2.5     | 127.03        | 141.4         |
| 2       | 42          | 3.2     | 66            | 81.41         |
| 3       | 53          | 3.1     | 58.32         | 67.87         |
| 4       | 39          | 2.8     | 80.8          | 90.5          |
| 5       | 33          | 2.9     | 83.4          | 99.3          |
| 6       | 68          | 2.7     | 70.9          | 75.8          |
| 7       | 47          | 3.3     | 90.8          | 131.6         |
| 8       | 58          | 2.5     | 102.3         | 110.1         |

CCA: Clivus-canal angle, OH: Odontoid height, Preop: Preoperative, Postop: Postoperative
decompression of the CVJ, usually with odontoidectomy performed, firstly, followed by the POCF (in different or single surgical times).\[13,27,29,36,48]\]

Of the anterior decompression techniques, the transoral approach to removing the odontoid peg was the classic route until recently.\[31,36\] Due to the height of the odontoid in patients with BI, as observed in the present series, where the OH average was 2.87 ± 0.3 cm above the Chamberlain’s line, it is often necessary to add extensions. These include the vertical splitting of the soft and hard palates, as well as the horizontal splitting of the maxilla (unilateral Le Fort I), with great associated morbidity.\[8,24,26,31\]

Since its anatomical description in 2002,\[1\] and clinical application in 2005,\[25\] endonasal endoscopic odontoidectomy has become more commonly used when the odontoid is located above the palatine line.\[14,15,32\] The meta-analysis that compared the complications of transoral and endonasal odontoidectomy found a statistically significant difference only in terms of the need for tracheostomy, which was greater after transoral odontoidectomy.\[35\] Hypothetical advantages of the endonasal technique still include reduction in the swelling of the tongue and oropharynx, and the nondivision of the soft palate, causing lower degrees of velopalatine dysfunctions and allowing the early start of oral feeding.\[14\]

Endonasal endoscopic odontoidectomy requires high expertise from the surgeon, as the working distance is quite long, especially in retroverted odontoids, and it is difficult to do it in obese patients and in the presence of associated subaxial deformities of the cervical spine.\[14,19,35\] The authors believe that due to the complex anatomical changes present in patients with BI of Type B of Goel, especially when there are associated rotational deformities, the use of transoperative neuronavigation is mandatory. Still in this group, due to the retroversion of the clivus that protrudes into the brainstem together with the odontoid, the authors advocate that for the decompression to be effective, it must incorporate the lower portion of the clivus. This greatly increases the complexity of the procedure due to the height at which this structure is positioned in these patients (personal communication).

The indirect reduction of the odontoid by the application of trans-skeletal external traction or by intraoperative maneuvers during POCF has been used in patients with associated AAD (Goel Type A), even in cases considered irreducible, thus eliminating the need for ventral approach to remove the odontoid peg.\[5,6,34,37\] In patients under 14 years of age, the indirect reduction of the odontoid through the posterior approach has also been described as successful.\[26,36\]

Kim et al. in 2004\[26\] used the same biomechanical principles applied in the present description for indirect odontoid decompression in patients with BI in the pediatric age group and found improvement in the CCA with statistical significance. However, they used a halo ring to perform the maneuvers.\[26\] More recently, Goel expanded the indication of posterior C1/C2 fixation alone to type B patients of his classification, however, with the caveat that there are few series that evaluate the treatment of this specific group of patients.\[20\] The degree of deformity was very accentuated in the present series, as observed by means of odontoid height and CCA, which makes the use of short fixations including only C1 and C2 difficult to perform, as highlighted by Goel himself, which describes high rates of complications even in experienced hands.\[6,20\] The authors recommend the use of subaxial segments, due to their more predictable anatomy, in patients with BI of Type B of Goel.

In the region of the current study, these patients with BI of Type B of Goel have an eversion into the posterior fossa of the constituent components of the margins of the foramen magnum. The deformity goes far beyond the simple insinuation of the odontoid through the foramen magnum. It is probable that fixation restricted to segments C1 and C2 is not able to reverse, even partially, the complex deformities present in these patients that include the clivus and occipital bone (personal communication).

The technique discussed here is simple and highly reproducible in any neurosurgery service without requiring major technological resources, except for a 3-point “Mayfield” type head clamp. All patients submitted to the present technique showed an increase in the CCA, a mean of 14.81 ± 1.54°, with a statistically significant difference between the pre and postoperative periods, as described by Kim et al. in children.\[26\] The fact that patients with BI have deformity kyphosis in the CVJ\[7\] with compensatory subaxial hyperlordosis also seems to benefit from the present technique since applied hyperextension acts on CVJ fulcrum, with no apparent accentuation on the cervical lordosis.\[26\] In fact, there was an improvement in the hyperlordosis associated with BI (personal communication). Due to the complexity of the changes mentioned, the authors believe that the inclusion of the occiput and the average use of three levels of the subaxial segment of the cervical spine (C3, C4, and C5) are necessary. All patients underwent posterior osteodural decompression of the CVJ without manipulation of the tonsils since they were associated with Type 1 Chiari malformation.\[20,26\] In addition, this bone space created makes it possible to accommodate CVJ in extension after the maneuver.\[6\]

The present manuscript is intended to describe a suitable technique for treating patients with BI of the “type B” of Goel associated with pronounced CVJ deformities, usually found in the study region. The lack of clinical outcomes analysis should be the focus of forthcoming analyses. Although not the subject of the present study, none of the patients required posterior decompression at a later stage. There was
an appreciable neurological improvement in all patients. The rate of complications was low and manageable.

Probably, ventral approaches for removing the odontoid peg will remain only as a rescue procedure in patients who underwent an attempt to indirect posterior decompression and who did not obtain a satisfactory clinical response associated with radiological evaluation showing persistence of brainstem compression.[37]

There were many limitations to this study, particularly the small number of patients and the lack of clinical data, especially for long-term follow-up. Since it was carried out in a public health system hospital in one of the poorest regions of Brazil (Northeast), there was no access to technologies such as intraoperative neurophysiological monitoring and transoperative image acquisition system and navigation, which have been shown to be beneficial.[23,26,42]

CONCLUSION

The indirect decompression of the brainstem by the odontoid peg during POCE, using distraction-extension maneuvers of the cephalic segment with the aid of the “Mayfield” head clamp in patients with BI of type B of Goel, seems to be a promising technique in this complex group of patients with multiple CVJ malformations. It has the advantage of single approach reduction and fixation, decreasing surgical morbidity. All patients in this series showed radiological improvement of the CCA and associated neurological improvement.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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