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ORIGINAL ARTICLE

Surgical outcomes of basilar invagination type B without atlantoaxial dislocation through simple posterior fossa decompression: a retrospective study of 18 cases

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Abstract

Background Basilar invagination (BI) is a common disease in the craniocervical junction (CVJ) area. Posterior fossa decompression with/without fixation is a controversial surgical strategy for BI type B. This study aimed to evaluate the efficacy of simple posterior fossa decompression in treating BI type B.

Methods This study retrospectively enrolled BI type B patients who underwent simple posterior fossa decompression at Huashan Hospital, Fudan University between 2014.12 and 2021.12. Patient data and images were recorded pre- and post-operatively (at the last follow-up) to evaluate the surgical outcomes and craniocervical stability.

Results A total of 18 BI type B patients (13 females), with a mean age of 44.2±7.9 years (range 37–62 years), were enrolled. The mean follow-up period was 47.7±20.6 months (range 10–81 months). All patients received simple posterior fossa decompression without any fixation. At the last follow-up, compared with preoperation, the JOA scores were significantly higher (14.2±1.5 vs. 9.9±2.0, $p = 0.001$); the CCA was improved (128.7±9.6° vs. 121.5±8.1° $p = 0.001$), and the DOCL was reduced (7.9±1.5 mm vs. 9.9±2.5 mm, $p = 0.001$). However, the follow-up and preoperative ADI, BAI, PR, and D/L ratio were similar. No patients had an unstable condition between the C1-2 facet joints that was observed in the follow-up CT and dynamic X-ray.

Conclusions In BI type B patients, simple posterior fossa decompression could improve neurological function and will not induce CVJ instability in BI type B patients. Simple posterior fossa decompression could be a satisfactory surgical strategy for BI type B patients, but preoperative CVJ stability assessment is crucial.

Key words BI type B · CVJ stability · Simple posterior fossa decompression

Abbreviations

ADI	atlantodental interval
AOJ	atlanto-occipital joint
BAI	basion-axis interval
BDI	basion-dens interval
BI	basilar invagination
CCA	the clivus-canal angle
CVJ	craniocervical junction
D/L ratio	the atlanto-occipital joint depth/length ratio
DOCL	the distance of the odontoid apex to Chamberlain's line

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O-C1 angle	atlanto-occipital angle
PFD	posterior fossa decompression
PFDD	posterior fossa decompression with duraplasty
PR	Powers ratio
SD	standard deviation

Introduction

Basilar invagination (BI) is a common ossification-malformation in the craniocervical junction (CVJ) area and is characterized by abnormal protrusion of the odontoid process to the foramen magnum, resulting in crowding in the foramen magnum and compression symptoms of the medulla oblongata, upper cervical spinal cord and posterior cranial nerves [28–30, 32, 38]. In recent years, a classification based on the stability of the CVJ was proposed [16, 23]: BI type A (type A BI, BI type I) is defined as an unstable basilar invagination that is combined with atlantoaxial dislocation or any other upper cervical spine instability factors; in contrast, BI type B (type B BI, BI type II) is regarded as a relatively stable basilar invagination without distinct upper cervical instability factors [9, 11, 17].

In clinical practice, for unstable BI type A treatment, the principles of reduction, decompression, fusion and fixation have been well recognized by neurosurgeons [40].

Among the surgical methods for BI type B, simple posterior decompression, such as posterior fossa decompression (PFD) or posterior fossa decompression with duraplasty (PFDD), is a common strategy since BI type B is relatively stable. Moreover, simple posterior decompression without any fixation has been demonstrated to be able to effectively relieve the compress of the oblongata and superior cervical medulla and achieve a satisfying prognosis with less surgical damage and more surgical safety [4, 10, 35]. However, recently, some neurosurgeons have argued that BI type B has potential instability. Therefore, simple posterior decompression is not enough, and CVJ fusion and fixation are the key steps in treating BI type B [20, 22].

As the surgical strategies have been widely debated, various kinds of surgeries for BI type B treatment have been performed. Therefore, we designed this study to evaluate the efficacy of simple posterior fossa decompression without any fixation in BI type B treatment. In this retrospective study, we detailed the clinical characteristics and surgical outcomes for 18 BI type B cases using simple posterior fossa decompression to add more information and evidence for BI type B treatment.

Methods and materials

Patient population and study design

A retrospective, observational study was performed. BI type B patients who were admitted to the Department of the Neurosurgery at Fudan University Huashan Hospital between Dec. 2014 and Dec. 2021 were included. This study was approved by the Ethical Review Boards of Fudan University Huashan Hospital. Informed consent was obtained from all individual participants. If a patient cannot sign informed consents by himself, informed consents would be signed by his statutory guardians. The article is reported following the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) reporting guidelines. All patients were evaluated and treated by full-time neurosurgeons with specific fellow training, and all surgeries were performed by two attending neurosurgeons.

Inclusion criteria included the following: (a) age from 18 to 65 years; (b) diagnosis of BI type B; (c) underwent simple posterior fossa decompression to treat BI.

Exclusion criteria included the following: (a) any instability factors evaluated by classical CVJ stability judgement (such as ADI, BAI, BDI) pre-opeartion; (b) underwent CVJ area or craniocerebral surgery previously; (c) combined other CVJ abnormities (such as CVJ tumors, CVJ trauma); (d) reject to participate the study.

Diagnosing criteria of BI type B

(a) The odontoid process above the Chamberlain line more than 5mm in sagittal computed tomography (CT) scan; (b) the atlantodental interval (ADI) was within the normal range, no more than 3mm in sagittal computed tomography (CT) scan and without any instable factors confirmed by dynamic X-ray or CT.

Radiological evaluation

CT scans of CVJ area (0.6 mm each slice) with the head in neutral were obtained pre- and post-operatively (at last follow-up) from each BI type B patient on a 256-slice multidetector CT scanner (Siemens, Germany) to evaluate the stability of CVJ area. The high-resolution T2-weighted magnetic resonance images of CVJ area were also obtained on a 3.0T GEMR750 MRI scanner with an eight-channel phased-array head coil (Siemens, Germany) to evaluate the level of magnum foramen compression induced by BI and the volume of intramedullary syringemyelia. Dynamic X-ray was also performed to evaluate CVJ instability.

To evaluate the basilar invagination and CVJ stability (atlantoaxial and atlanto-occipital stability) pre- and post-operatively (at last follow-up), the clivus-canal angle (CCA) (Fig. 1a), the distance of the odontoid apex to Chamberlain's line (DOCL) (Fig. 1b), the atlantodental interval (ADI) (Fig. 1c), the basion-dens interval (BDI) (Fig. 1e), the basion-axial interval (BAI) (Fig. 1f), the Powers ratio (PR) (Fig. 1g), and the atlanto-occipital angle (O-C1 angle) (Fig. 1i) on sagittal median CT were measured. In addition, the atlanto-occipital joint depth/length ratio (D/L ratio) (Fig. 1h) on paramedian sagittal CT and the position of C1-2 facets joints (Fig. 1d) in coronal CT were also evaluated (Table 1) [2, 3, 7, 12, 15, 24, 25, 27, 39, 41].

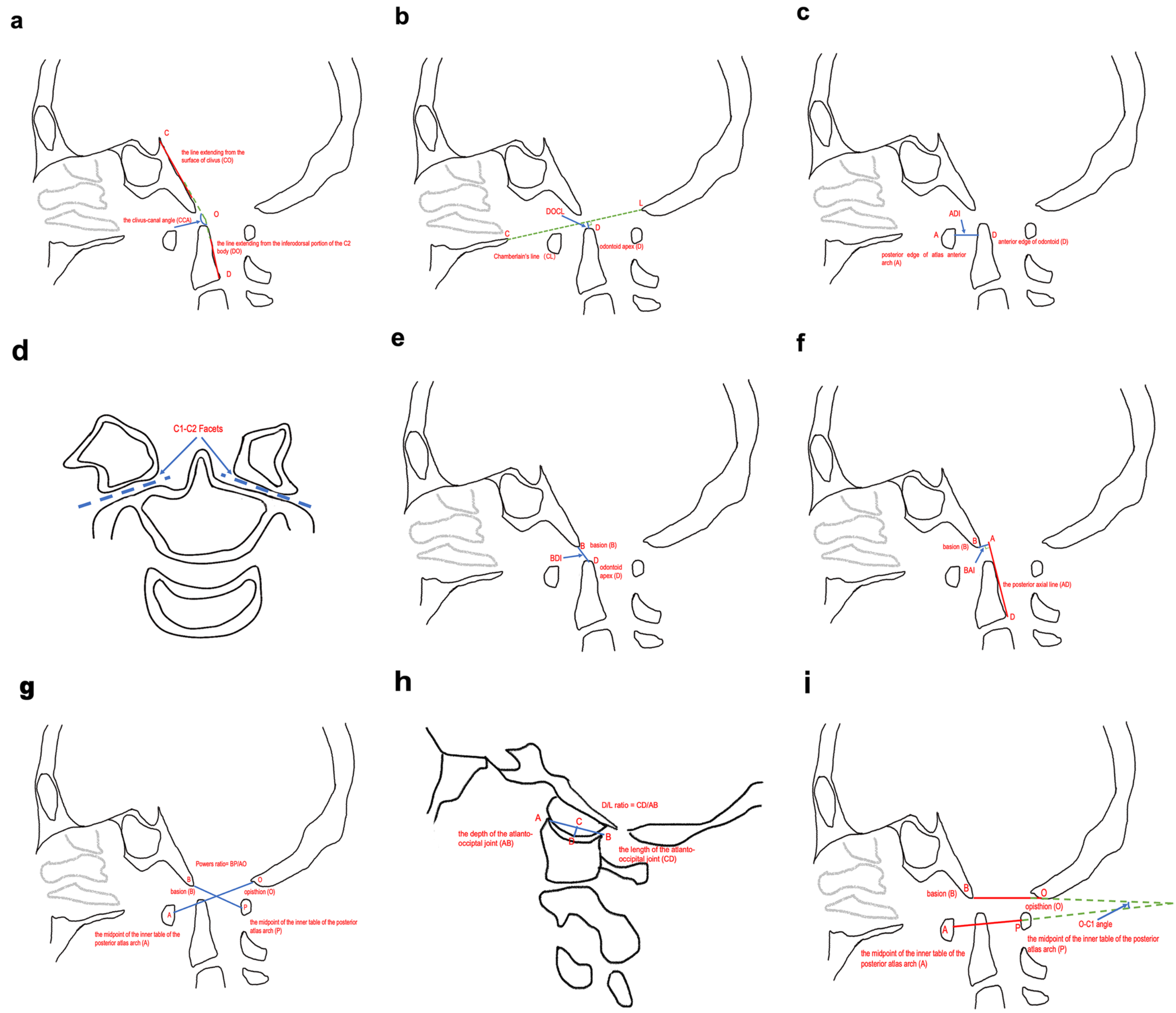

Fig. 1 Diagrams of CVJ area measurement on mid-sagittal, paramedial-sagittal, and coronal images**. a** The line extending from the surface of the clivus (CO), the line extending from the inferodorsal portion of the C2 body (DO), and the angle (COD) was the clivus-canal angle (CCA). **b** Chamberlain's line (CL), odontoid apex (D), the distance from point D to the line CL was the distance from the odontoid apex to Chamberlain's line (DOCL). **c** Posterior edge of the atlas anterior arch (A), anterior edge of the odontoid (D), and the length of segment AD was the atlantodental interval (ADI). **d** The blue dotted line indicates the relationship between the position of two atlantoaxial lateral mass articular facets, which were used to evaluate the stability of the atlantoaxial. **e** Basion (B), odontoid apex (D), the length of BD was the basion-dens interval (BDI). **f** Basion (B), the posterior axial line (AD), the distance from the basion (B) to the posterior axial line (AD) was the basion-axis interval (BAI). **g** Basion (B), opisthion (O), the midpoint of the inner table of the posterior atlas arch (A), and the midpoint of the inner table of the posterior atlas arch (P). The ratio of BP to AO was the powers ratio (PR). **h** The depth of the atlanto-occipital joint (AB), the length of the atlanto-occipital joint (CD), and the ratio of CD to AB were calculated as the atlanto-occipital joint depth/length ratio (D/L ratio). **i** Basion (B), opisthion (O), the midpoint of the inner table of the posterior atlas arch (A), and the midpoint of the inner table of the posterior atlas arch (P). The angle (O-C1) formed by the extension line along AP and the extension line along BO was the atlanto-occipital angle (O-C1 angle)

Table 1 Definition of radiological evaluation indexes for BI and CVJ stability

Abbreviation	Full form	Definition
ADI	Atlantodental interval	Distance from the posterior edge of atlas anterior arch to the anterior edge of odontoid
BAI	Basion-axis interval	Distance from the basion to the tangential line at the posterior surface of the dens
BDI	Basion-dens interval	Distance from the basion to the tip of the dens
CCA	The clivus-canal angle	Angle formed by the line along the posterior edge of the odontoid and the line along the inferior clivus
C1-2 facets	The position of C1-2 lateral facets	Relationship between the position of two atlantoaxial lateral masses articular facets
D/L ratio	The atlanto-occipital joint depth/length ratio	Ratio of the depth of the atlanto-occipital joint to the length of the atlanto-occipital joint
DOCL	The distance of the odontoid apex to Chamberlain's line	Distance from the odontoid apex above the Chamberlain's line
O-C1 angle	Atlanto-occipital angle	Angle formed by the line between the anterior and posterior borders of the foramen magnum and the line through the centers of the anterior and posterior arches of the atlas
PR	Powers ratio	Powers ratio was calculated by dividing the distance between the occipital base (O) and the posterior of the anterior arch of C1 (A) by the distance between the caudal aspect of the basion (B) and the spinolaminar line (P); Powers ratio = BP/AO

Neurological function evaluation

The Japanese Orthopaedic Association (JOA, version 1975) Score [14, 42] was used to evaluate neurological function of each patient enrolled pre- and postoperatively (at last follow-up), performed by at least two qualified independent investigators in a blinded manner.

Surgical strategies

PFD (posterior fossa decompression) or PFDD (posterior fossa decompression with duraplasty) as a simple posterior fossa decompression procedure was performed in previous studies [1, 5]. For PFD, through the posterior midline approach, a bone-window suboccipital craniectomy (diameter, 3–4 cm) was performed, the posterior arch of the atlas (1.5–2 cm) was resected, and the ligaments, soft tissues and other structures attached to the dura were also removed to achieve complete decompression of the magnum foramen. For PFDD, a dural incision was subsequently performed, and the attached arachnoid membrane, especially around the medial and lateral apertures of the fourth ventricle, was thoroughly released. Then, the dura mater was grafted with the autologous graft. The wound was closed in layers.

Statistical analysis

Continuous variables were expressed as means ± standard deviation (SD) or medians (interquartile range) and categorical variables as percentages. The univariate analyses of categorical data were performed using the chi-squared test. Equality of variance was assessed using Levene's test. Normally distributed variables were compared using Student's t-tests or one-way ANOVA, whereas non-normally distributed variables were compared using the Kruskal-Wallis or Mann–Whitney U-tests. All statistical tests were two-tailed and p values < 0.05 were considered statistically significant. Statistical analysis was carried out using IBM SPSS Statistics (version 26, IBM, U.SA.).

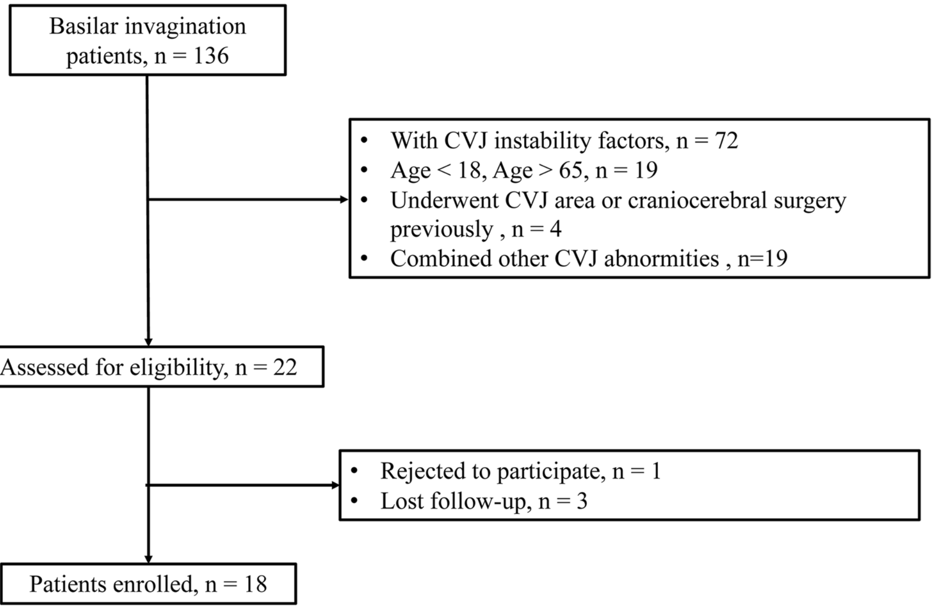

Fig. 2 Flowchart depicting our study cohort

Table 2 Demographic and baseline clinical characteristics among 18 type B BI patients

Characteristics (*N*=18)	Number
Mean age, year±SD	44.2±7.9
Male, *n* (%)	5 (27.8)
Female, *n* (%)	13 (72.2)
Follow-up, mean±SD	47.7±20.6
Symptoms	
Muscle weakness, *n* (%)	11 (61.1)
Limb numbness, *n* (%)	15 (83.3)
Ataxia, *n* (%)	5 (27.8)
Neck pain and motor restriction, *n* (%)	13 (72.2)
Dysphagia and dyspnea, *n* (%)	4 (22.2)
Comorbidities	
Atlantooccipital fusion, *n* (%)	8 (44.4)
C2-3 fusion, *n* (%)	4 (22.2)
Syringomyelia, *n* (%)	13 (72.2)
Surgical plan	
PFDD, *n* (%)	13 (72.2)
PFD, *n* (%)	5 (27.8)

N number of patients, *SD* standard deviation

Results

Patient characteristics

A total of 18 patients with BI type B (13 females) with a mean age of 44.2±7.9 years (range 37-62 years) were included. The mean follow-up period was 47.7±20.6 months (range 10-81 months). All patients received simple posterior fossa decompression without any fixation (Fig. 2) (Table 2).

All patients presented with one or more neurological symptoms. Eleven (61.1%) patients presented with muscle weakness, 15 (83.3%) with limb numbness, 5 (27.8%) with ataxia, 13 (72.2%) with neck pain and mobility restriction and 4 (22.2%) with dysphagia or dyspnoea. Moreover, 8 (44.4%) patients had atlantooccipital fusion, 4 (22.2%) had C2-3 fusion, and 13 (72.2%) had syringomyelia (Table 3).

Decompression of the posterior fossa was favourable in BI type B patients

To assess the decompression efficiency, the CCA and DOCL obtained from CT scans were used. Compared with preoperation, the CCA at the last follow-up significantly increased (128.7±9.6° vs. 121.5±8.1°, $p < 0.001$); however, the DOCL at the last follow-up was reduced (7.9±1.5 mm vs. 9.9±2.5 mm, $p < 0.001$).

In addition, compression of the ventral dural sac was decreased in 17 of the BI type B patients (17/18, 94.4%), and the volume of syringomyelia was reduced in 12 patients (12/13, 92.3%) (Fig. 3) (Table 4). Only one patient who had suffered progressed syringomyelia also showed continuous compression to the ventral medulla, indicating that sufficient decompression was favourable in BI type B patients.

Simple posterior fossa decompression was associated with neurological functional improvements in BI type B patients

At the last follow-up, the postoperative JOA scores of all patients were significantly higher than the preoperative scores ($p < 0.001$)

Table 3 Baseline and clinical characteristic of each BI type B patient

Number	Gender	Age	Follow-up (m)	Surgical strategy	Symptom and comorbidities[a]							
					Muscle weakness	Limb numbness	Ataxia	Neck pain Motor restriction	Dysphagia Dyspnea	Atlantooccipital fusion	C2-3 fusion	Syringomyelia
1	Female	53	80	PFDD	−	+	−	+	−	+	−	+
2	Female	41	81	PFDD	+	+	−	+	+	−	+	+
3	Male	41	73	PFD	-	-	−	+	−	+	−	+
4	Female	38	58	PFDD	+	+	+	+	−	−	−	+
5	Female	43	62	PFD	-	+	+	−	−	+	−	−
6	Female	40	35	PFDD	+	+	−	+	−	+	−	+
7	Female	38	41	PFDD	+	+	−	−	−	−	+	−
8	Female	42	37	PFDD	+	+	−	+	−	+	−	+
9	Female	51	62	PFDD	+	+	−	+	−	−	+	+
10	Male	29	60	PFD	-	+	−	−	−	−	−	−
11	Female	54	50	PFDD	-	+	−	−	+	−	+	+
12	Male	42	54	PFDD	+	+	+	+	−	+	−	+
13	Female	45	42	PFDD	-	-	−	+	−	−	−	+
14	Male	51	37	PFD	-	+	−	+	−	+	−	−
15	Female	62	40	PFDD	+	+	+	+	+	+	−	+
16	Female	38	14	PFDD	+	+	−	−	−	−	−	+
17	Female	37	22	PFDD	+	+	−	+	−	−	−	−
18	Female	50	10	PFD	+	-	+	−	+	−	−	+
Mean±SD		44.2±7.9	47.7±20.6									

[a]"+" means with this symptom or comorbidities, "−" means without such symptom or comorbidities

Data were given as Mean±SD unless otherwise noted. *SD* standard deviation, *PFDD* posterior fossa decompression with duraplasty, *PFD* posterior fossa decompression

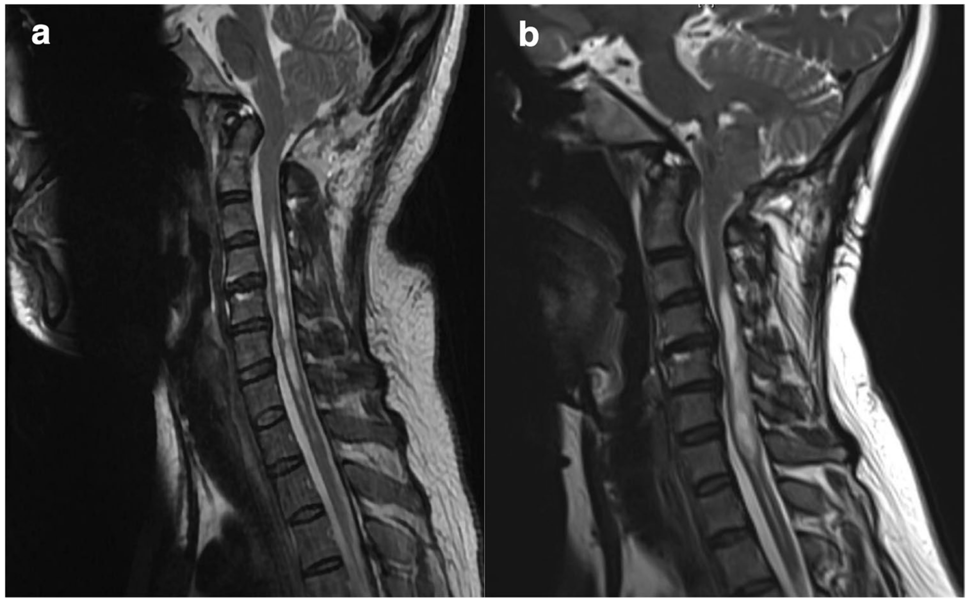

Fig. 3 Image of decompression effects on a mid-sagittal position at last follow-up. **a** Mid-sagittal image of T2 MR showed syringomyelia and the compression level of the ventral dural sac of the medulla oblongata with abnormal CCA and DOCL in type B BI postoperatively. **b** Mid-sagittal T2 MR image showing syringomyelia and the compression level of the ventral dural sac of the medulla oblongata with improved CCA and DOCL in Type B BI at follow-up

Table 4 Evaluation of surgical effects for each BI type B patient in post-operative (at last follow up)

Number	JOA sores		CCA (°)		DOCL (mm)		Ventral brainstem compression[a]		Syringomyelia[a]	
	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u
1	8	13	115	133	11.8	7.3	+	i	+	i
2	9	15	135	144	10.0	7.1	+	i	+	i
3	10	15	108	121	9.1	8.1	+	i	+	i
4	9	10	132	146	9.0	8.2	+	+	+	+
5	9	14	115	120	13.1	9.5	+	i	+	−
6	10	14	118	116	14.3	10.3	+	i	−	−
7	7	12	123	127	12.6	7.5	+	i	+	−
8	8	14	121	121	9.6	6.8	+	i	−	−
9	10	14	120	126	13.8	10.4	+	i	+	i
10	11	14	130	131	10.2	8.4	+	i	+	i
11	8	15	115	127	8.7	7.4	+	i	−	−
12	7	15	109	120	7.3	7.1	+	i	+	i
13	13	14	125	127	8.4	7.0	+	i	+	i
14	12	14	127	130	10.1	7.9	+	i	+	i
15	11	15	114	118	11.2	10.5	+	i	−	−
16	14	17	133	140	7.0	5.6	+	i	+	i
17	12	16	119	124	6.6	6.4	+	i	−	−
18	11	15	127	146	5.7	6.1	+	i	+	i
Mean±SD	9.9±2.0	14.2±1.5	121.5±8.1	128.7±9.6	9.9±2.5	7.9±1.5				
p value	<0.001*		<0.001*		<0.001*					

*$p<0.05$

[a] "+" means with this symptom or comorbidities, "−" means without such symptom or comorbidity or completely curing it; "i" means the symptom or comorbidity improved

Data were given as Mean±SD unless otherwise noted. *SD*, standard deviation; *CCA*, clivus-canal angle; *DOCL*, the odontoid apex to Chamberlain's line; *pre op*, pre-operation, *f/u* follow-up

Table 5 Pre- and post-operative symptoms and comorbidities conditions of each BI type B patient

Number	Symptom and comorbidities[a]									
	Muscle weakness		Limb numbness		Ataxia		Neck pain Motor restriction		Dysphagia Dyspnea	
	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u
1	−	−	+	i	−	−	+	i	−	−
2	+	i	+	−	−	−	+	i	+	−
3	−	−	−	−	−	−	+	−	−	−
4	+	+	+	+	+	+	+	+	−	−
5	−	−	+	i	+	−	−	−	−	−
6	+	i	+	i	−	−	+	−	−	−
7	+	i	+	i	−	−	−	−	−	−
8	+	i	+	i	−	−	+	i	−	−
9	+	i	+	i	−	−	+	−	−	−
10	−	−	+	i	−	−	−	−	−	−
11	−	−	+	i	−	−	−	−	+	−
12	+	−	+	i	+	−	+	i	−	−
13	−	−	−	−	−	−	+	i	−	−
14	−	−	+	i	−	−	+	i	−	−
15	+	−	+	−	+	−	+	−	+	−
16	+	−	+	−	−	−	−	−	−	−
17	+	i	+	−	−	−	+	i	−	−
18	+	i	−	−	+	−	−	−	+	−

[a] "+" means with this symptom or comorbidities, "−" means without such symptom or comorbidity or completely curing it; "i" means the symptom or comorbidity improved

Pre op pre−operation, *f/u* follow−up

(Table 4). Ten patients (out of 11, 90.9%) showed improvement in muscle strength, with three of them recovering fully. Limb numbness was alleviated in 14 patients (out of 15, 93.3%), with four of them recovering fully. Four out of five patients (80%) recovered from ataxia, whereas 11 out of 12 patients (91.7%) experienced improvement in neck pain and mobility restriction, with four of them recovering fully. All patients who suffered from dysphagia and dyspnoea recovered (Table 5).

Our study showed that in most BI type B patients, simple posterior fossa decompression was associated with improvement in neurological functions, with 94.4% (17/18) of the participants achieving distinct improvement in their neurological symptoms. However, one patient (patient #4) did not show significant neurofunction improvement after simple posterior fossa decompression, which we presumed was due to insufficient decompression. These results suggest that satisfactory decompression by simple posterior decompression may be associated with neurological outcome improvements in BI type B patients.

Simple posterior fossa decompression was not associated with CVJ instability in BI type B patients

In general, CVJ stability includes bilateral atlantoaxial and atlanto-occipital stability. For atlantoaxial stability evaluation, no differences in the ADI were found pre- and postoperation (1.5±0.3 mm vs. 1.6±0.4 mm, $p = 0.178$). Additionally, the ADI of each BI type B patient, whether measured pre- or postoperatively (at last follow-up), was within the normal range (Fig. 4a, b). Moreover, in dynamic X-ray, normal physiological relationships of the C1-2 facet joint positions were observed in all patients at the last follow-up (Table 6).

In addition, BDI, BAI, PR, D/L ratio, and O-C1 angle were used to assess atlanto-occipital stability. Our results showed that no differences in BAI, PR or D/L ratio were observed pre- and postoperation (6.2±2.2 vs. 5.9±2.3 mm, $p = 0.064$; 0.70±0.08 vs. 0.73±0.05 $p = 0.063$; 0.27±0.06 vs. 0.24±0.05, $p = 0.055$, respectively). Moreover, the BAI, PR and D/L ratio, either measured pre- or postoperatively (at the last follow-up), were within the normal range. Similarly, although the BDI and O-C1 angle had statistically significant differences pre- and postoperation ($p = 0.036$; $p = 0.001$), the pre- and postoperative BDI and O-C1 angle in all patients were within the normal range (6.9±1.9 vs. 6.5±2.1 mm; 5.0±1.3° vs. 3.6±1.7°). According to previous literature, a BDI > 12 mm or an O-C1 angle far from the neutral position (5.06 ±0.48°) was considered atlanto-occipital dislocation [33]. Therefore, our data indicated that no distinct CVJ instability was observed after simple fossa decompression (Fig. 4c, d, e) (Table 7).

In summary, no progressive CVJ instability was detected after simple posterior fossa decompression in any of the included BI patients.

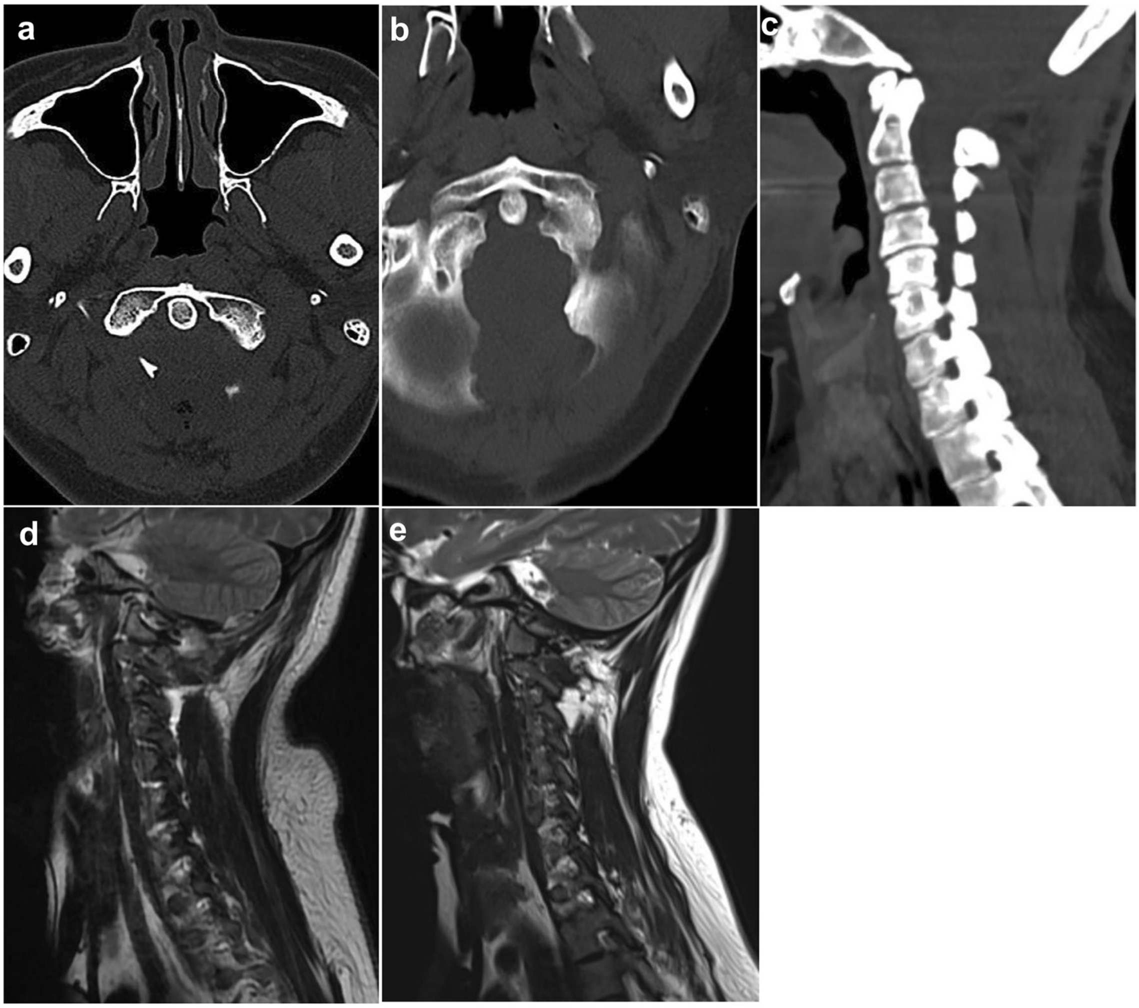

Fig. 4 Images of CVJ stability in mid-sagittal and horizontal positions at the last follow-up. **a** Horizontal CT scan showing type B BI with normal ADI postoperatively. **b** Horizontal CT scan confirmed normal postoperative ADI without any deterioration at the last follow-up. **c** Mid-sagittal CT scan showing confirmed normal postoperative BDI and BAI without any deterioration at the last follow-up. **d** Paramedial-sagittal T2 MR image showing the condition of the atlanto-occipital joint with a normal depth/length ratio postoperatively. **e** Paramedial-sagittal T2 MR image showing the natural position of the atlanto-occipital joint facets with a normal depth/length ratio at the last follow-up

Discussion

In this study, we found that simple posterior fossa decompression could effectively decrease the compression of the ventral medulla oblongata and improve neurological function in BI type B patients. Moreover, simple posterior fossa decompression will not induce CVJ instability in BI type B patients.

Posterior fossa decompression may improve neurological outcomes in BI type B patients

"Decompression" is the classical surgical procedure for BI and has been performed since 1968 [4]. Posterior fossa decompression could achieve satisfactory outcomes in some BI cases [6, 10, 31]. However, in BI patients with complicated conditions of CVJ stability, especially in BI type B patients, the choice of surgical strategy is controversial. Some scholars believe that due to the potential instability in BI type B, fusion and fixation are able to improve outcomes in these patients [18, 20, 22, 43]. However, fusion and fixation procedures do not seem to be a "wonderful" procedure without risks. For instance, fusion and fixation might injure the C1-2 nerve root, spinal cord, and vertebral artery and limit the range of movements of the cervical spine. Moreover, fusion and fixation were considered to be associated with a longer duration of surgery, higher cost, more blood loss, and implant risks [36, 37]. Our study found that PFD/PFDD could improve neurological outcomes in BI type B patients, indicating that simple posterior fossa decompression may be sufficient for BI type B patients whose preoperative CVJ stability is favourable. We suggest that not all BI type B patients require fusion and fixation

Table 6 Evaluation of pre- and post-operative atlantoaxial stability in each BI type B patient

Number	ADI (mm)		C1−2 facets conditions in CT[a]		C1−2 facets conditions in dynamic X ray[b]	
	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u
1	1.9	1.6	Normal	Normal	Normal	Normal
2	1.5	1.1	Normal	Normal	Normal	Normal
3	1.0	1.2	Normal	Normal	Normal	Normal
4	1.5	1.8	Normal	Normal	Normal	Normal
5	1.8	1.8	Normal	Normal	Normal	Normal
6	1.1	1.3	Normal	Normal	Normal	Normal
7	1.3	1.5	Normal	Normal	Normal	Normal
8	1.1	1.3	Normal	Normal	Normal	Normal
9	1.5	1.2	Normal	Normal	Normal	Normal
10	1.8	2.1	Normal	Normal	Normal	Normal
11	1.2	2.1	Normal	Normal	Normal	Normal
12	1.5	1.9	Normal	Normal	Normal	Normal
13	1.7	1.8	Normal	Normal	Normal	Normal
14	1.1	1.5	Normal	Normal	Normal	Normal
15	1.9	1.3	Normal	Normal	Normal	Normal
16	1.2	1.1	Normal	Normal	Normal	Normal
17	2.1	2.1	Normal	Normal	Normal	Normal
18	1.3	2.1	Normal	Normal	Normal	Normal
Mean±SD	1.5±0.3	1.6±0.4				
p value	$p = 0.178$					

[a] C1−2 facets conditions: the position of C1−2 lateral facets.

[b] Dynamic X ray: including the extension and flexion positions in X ray.

Data were given as Mean±SD unless otherwise noted. *SD*, standard deviation; *ADI,* atlantodental interval; *Pre op*, pre−operation; *f/u*, follow−up

and that the choice of fusion and fixation procedure for BI type B patients should be carefully considered.

Posterior fossa decompression and CVJ stability: preoperative stability assessment is crucial

To date, surgery has remained the most effective treatment for BI [38, 40]; however, it was previously reported that preoperative CVJ instability may cause neurological function deterioration after simple decompression [40]. This indicates that detailed assessments of CVJ stability should be carried out preoperatively.

Unlike BI type A, which is characterized by CVJ instability [13, 34, 38], it may not be necessary for BI type B patients to receive fixation and fusion. As previously mentioned, CVJ instability may worsen neurological outcomes in BI patients; thus, it was reasonable to assess the stability of CVJ preoperatively. We proposed that the therapeutic strategy of BI type B patients should be designed according to CVJ stability.

Additionally, our study aims to determine whether simple posterior decompression without fusion and fixation would affect the CVJ stability of BI type B patients at the last follow-up. In our study, atlantoaxial stability was assessed by the ADI and C1-2 facet positions, while the BDI, BAI, PR, O-C1 angle and atlanto-occipital D/L ratio were used to evaluate atlanto-occipital stability. Based on these indicators both pre- and postoperation, no CVJ instability was found in our patients' pre/post-simple posterior fossa decompression. Moreover, almost all follow-up patients achieved a certain degree of neurological improvement, indicating that simple fossa decompression may not harm CVJ stability in BI type B patients. However, we stress that if CVJ instability is identified preoperatively, other strategies, including fusion and fixation, may be needed.

Most favorable strategies to assess CVJ stability are unclear

For the evaluation of CVJ stability, the presence of atlantoaxial dislocation is a crucial factor. ADI and C1-2 facet position were the classical indices used to assess atlantoaxial stability. However, Goel et al. defined a new classification for atlantoaxial dislocation [18] and suggested that potential atlantoaxial instability existed in BI type B patients [19, 21, 43]; thus, "fusion and fixation" was

Table 7 Evaluation of pre- and post-operative atlanto-occipital stability in each BI type B patient

Number	BDI (mm)		BAI (mm)		PR		D/L ratio		O-C1 angle (°)	
	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u	Pre op	Last f/u
1	6.5	7.0	4.3	5.1	0.70	0.71	0.20	0.21	2.1	5.2
2	4.3	4.1	7.2	6.4	0.70	0.67	0.20	0.25	3.9	6.2
3	6.4	5.8	7.0	7.0	0.74	0.68	0.35	0.31	6.5	7.9
4	8.2	8.0	2.3	3.4	0.71	0.66	0.34	0.30	6.0	5.8
5	6.6	6.5	9.5	9.7	0.77	0.72	0.21	0.22	3.1	4.2
6	3.2	5.4	3.9	5.1	0.67	0.65	0.22	0.20	4.7	5.7
7	7.2	8.1	5.3	6.2	0.67	0.62	0.24	0.34	2.7	5.8
8	9.0	8.8	10.0	9.3	0.78	0.70	0.28	0.36	4.2	4.5
9	9.1	9.1	5.6	6.7	0.78	0.78	0.15	0.26	2.0	5.2
10	8.7	9.3	3.8	4.2	0.70	0.72	0.20	0.20	2.1	3.0
11	4.6	6.0	2.4	3.2	0.79	0.85	0.24	0.34	1.2	3.3
12	4.4	4.5	6.4	5.7	0.77	0.80	0.24	0.25	5.5	7.2
13	8.2	7.0	6.7	5.4	0.66	0.77	0.28	0.38	6.3	5.4
14	7.4	7.9	5.6	6.3	0.64	0.59	0.21	0.25	4.1	3.9
15	4.3	5.4	8.6	9.2	0.73	0.71	0.29	0.33	3.2	5.8
16	9.5	10.9	8.7	9.7	0.79	0.62	0.26	0.23	1.7	3.7
17	3.4	4.5	3.3	2.9	0.69	0.55	0.25	0.25	1.8	4.8
18	5.3	6.0	5.3	4.8	0.81	0.74	0.24	0.19	3.4	3.6
Mean±SD	6.5±2.1	6.9±1.9	5.9±2.3	6.2±2.2	0.73±0.05	0.70±0.08	0.24±0.05	0.27±0.06	3.6±1.7	5.0±1.3
p value	0.036*		0.064		0.063		0.055		< 0.001*	

*$p<0.05$

Data were given as Mean±SD unless otherwise noted. *SD* standard deviation, *BDI* basion-dens interval, *BAI* basion-axis interval, *PR* powers ratio, *D/L ratio* the atlanto-occipital joint depth/length ratio, *O-C1 angle* atlanto-occipital angle, *pre op* pre-operation, *f/u* follow-up

necessary in treating these patients [18, 20, 22]. However, to date, clinical evidence of "chronic instability," as described by Goel, is rare [40].

On the other hand, atlanto-occipital instability is also vital to CVJ stability. Lu et al. evaluated CVJ stability by measuring clivus length, clivo-canal angle (CCA), basion-dens interval (BDI), basion-axial interval (BAI), and other indicators in kinematic CT scans. Based on these indicators, they suggested that occipitocervical fusion, combined with foramen magnum decompression, was beneficial to BI type B patients [43]. However, in Lu et al.'s study, the index values, especially the classical atlanto-occipital stability indices (BDI, BAI), were mostly within the normal range. This may indicate that there was no obvious CVJ instability [25, 33]. Moreover, the relationship between joint range of motion and joint instability is not clear. Recently, Lu et al. provided a new classification of the atlanto-occipital joint (AOJ) by measuring the curvature of the AOJ, indicating that most Chiari malformation with BI type B may be classified as type III AOJ. Type III AOJ was considered to be atlanto-occipital instability [26].

In our study, similar to the above indices, we also focused on the atlanto-occipital joint depth/length (D/L ratio, similar to the curvature of the AOJ). However, as previously mentioned, we found no difference in the D/L ratio between pre- and postoperative BI type B patients. This may indicate that no significant AOJ stability depravation was observed in BI type B patients after simple fossa decompression.

To date, the diagnostic criteria of atlantoaxial and atlanto-occipital instability remain controversial [8, 12, 17, 23, 25, 33, 43]. Recently, more studies have paid more attention to the CVJ stability evaluation methods [8, 25, 39, 40], but more clinical evidence is required to verify these methods.

Limitations

There were several drawbacks in the current study. First, the sample size was relatively small. Second, in our future study, we will select dynamic CT scans, which are considered to be more sensitive and accurate for CVJ instability diagnosis than dynamic X-rays [43], to assess the CVJ stability of our patients. Therefore, caution should be exercised in interpreting our conclusions, and a prospective multicentre study is justified to further elucidate the potential mechanism of this phenotype.

Conclusion

In this study, we found that simple posterior fossa decompression could effectively decrease compression on the ventral medulla oblongata and improve neurological function in BI type B patients. Furthermore, simple posterior fossa decompression will not induce CVJ instability in BI type B patients. Therefore, simple posterior fossa decompression could be a satisfactory surgical strategy for BI type B patients, but preoperative CVJ stability assessment is crucial.

Author contributions The study was designed by Xing-Yu Chen. Data collection was performed by Wei Chen, Jian-Lan Zhao, Hao-Ru Dong, Long-Nian Zhou, Xiao Xiao and Gong Chen. Data analysis was performed by Xing-Yu Chen, Wei Chen, Jian-Lan Zhao, Hao-Ru Dong, Long-Nian Zhou, Xiao Xiao, Gong Chen. The first draft of the manuscript was written by Xing-Yu Chen. Xiao-Ming Che and Rong Xie supervised the study and revised the manuscript. All authors read and approved the final manuscript.

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Declarations

Ethical approval This study was approved by the Ethical Review Boards of Fudan University Huashan Hospital. The article is reported following the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) reporting guidelines. All patients were evaluated and treated by full-time neurosurgeons with specific fellow training, and all surgeries were performed by two attending neurosurgeons.

Informed consent Informed consent was obtained from all individual participants included in the study. If a patient cannot sign informed consents by himself, informed consents would be signed by his statutory guardians.

Conflict of Interest The authors declare no competing interests.

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