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Effectiveness of C1 Laminectomy for Chiari Malformation Type 1: Posterior Fossa Volume Expansion and Syrinx-Volume Decrease Rate

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Purpose: This study aimed to analyze the effect of foramen magnum decompression with C1 laminectomy (C1L) for Chiari malformation type 1 (CM-1) in terms of improving clinical symptoms, expanding posterior fossa volume, and decreasing syrinx volume.
Materials and Methods: Between January 2007 and June 2019, 107 patients with CM-1 were included. The median patient age was 13±13 years (range: 9 months–60 years), female-to-male ratio was 1:1, and average length of tonsil herniation was 13±5 mm (range: 5–24 mm). Surgical techniques were divided into four groups based on duraplasty or C1L usage. Among the study subjects, 38 patients underwent duraplasty and had their syrinx volumes measured separately on serial magnetic resonance imaging. A three-dimensional visualization software was used to evaluate the syrinx-volume decrease rate.
Results: Bony decompression exhibited a mere 20% volume expansion of the lower-half posterior fossa. C1L offered a 3% additional volume expansion, which rose to 5% when duraplasty was added (p=0.029). There were no significant differences in complication rate when C1L was combined with duraplasty (p=0.526). Syrinx volumes were analyzed in 38 patients who had undergone duraplasty. Among them, 28 patients who had undergone duraplasty without C1L demonstrated a 5.9% monthly decrease in syrinx volume, which was 7.5% in the remaining 10 patients with C1L (p=0.040).
Conclusion: C1L was effective in increasing posterior fossa volume expansion, both with and without duraplasty. A more rapid decrease in syrinx volume occurred when C1L was combined with duraplasty.

Key Words: Chiari, syrinx, duraplasty, foramen magnum decompression

INTRODUCTION

Chiari malformation type 1 (CM-1) was originally defined by Hans Chiari as an "elongation of the tonsils and the medial parts of the inferior lobes of the cerebellum into cone-shaped projections which accompany the medulla oblongata into the spinal canal".[1] Today, it is typically defined as a cerebellar tonsil herniation ≥5 mm below the foramen magnum.[2-5] CM-1 is significantly associated with the formation of spinal cord syrinxes.[6-15]

CM-1 is treated using foramen magnum decompression (FMD), which relieves its symptoms by restoring cerebrospinal fluid (CSF) circulation at the foramen magnum.[16] The extent of FMD and necessity to resect a portion of the posterior arch of C1 and perform duraplasty, dural splitting, peeling, and dural closure remain debatable.[17-26]

FMD without duraplasty does not cause postoperative CSF leakage; however, it is potentially associated with an increased reoperation rate due to insufficient posterior fossa decompression and CSF circulation. Surgery with duraplasty is more efficient in increasing volume expansion and CSF circulation.[19] Nevertheless, reducing postoperative complications and inserting allogenic material, among other procedures, remain

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challenging. Incorporating duraplasty yields significant differences among surgical techniques; however, the inclusion of C1 laminectomy (C1L) is less challenging in that it nullifies increased CSF leakage and minimizes differences across surgical techniques. C1L potentially maximizes posterior fossa expansion without inducing CSF leakage.

Certain studies have revealed that treatment response is significantly influenced by the degree of posterior fossa volume increase.[27,28] Therefore, this study essentially measured the posterior fossa area expansion rate to evaluate surgical outcomes. The surgical techniques were divided into four groups according to duraplasty and C1L usage. The rate of syrinx-volume decrease was also analyzed to evaluate the improvement in CSF circulation at the foramen magnum level.

MATERIALS AND METHODS

Between January 2007 and June 2019, 107 patients with symptomatic CM-1 underwent FMD. The median age was 13±13 years (range: 9 months–60 years), male-to-female ratio was 1:1, and average length of tonsil herniation was 13±5 mm (range: 5–24 mm). The mean follow-up period was 42±29 months. Clinical symptoms included headache and dizziness (n=32, 30%), dysesthetic pain or weakness (n=34, 32%), ataxia or unsteadiness (n=7, 7%), scoliosis (n=22, 21%), cranial nerve dysfunction (n=4, 4%), and drop attacks (n=3, 3%). Sixty-eight patients (64%) had syringomyelia, and eight patients (8%) had hydrocephalus (Table 1).

Patients were categorized into four groups based on the surgical method. Group A-1 underwent only bony decompression (n=6) and C1L was added on group A-2 (n=21). Both groups B-1 (n=55) and B-2 (n=25) included patients who underwent duraplasty; however, group B-2 patients also under-

Table 1. Patient Characteristics and Clinical Symptoms (n=107)

Characteristics	Value
Median age (yr)	13±13 (range: 9 months–60 years)
Male-to-female ratio	1:1
Follow-up period (months)	42±29
Tonsil herniation (mm)	13±5 (range: 5–24)
Lower margin of tonsil	
Foramen magnum–C1	29 (27)
C1	34 (32)
C1–C2	32 (30)
C2	12 (11)
Clinical symptoms	
Headache or dizziness	32 (30)
Dysesthetic pain or weakness	34 (32)
Ataxia or unsteadiness	7 (7)
Scoliosis	22 (21)
Cranial nerve dysfunction	4 (4)
Drop attacks	3 (3)
Presentation	
Syringomyelia	68 (64)
Hydrocephalus	8 (8)

Data are presented as mean±standard deviation or n (%).

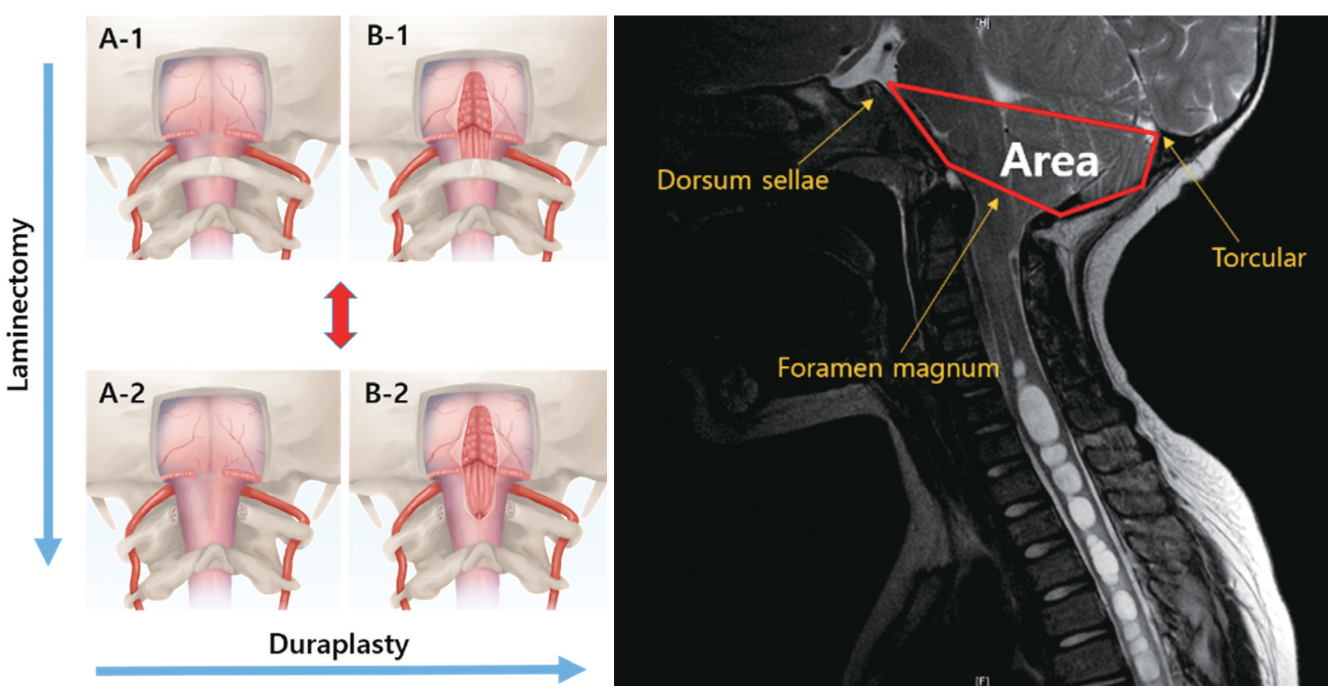

Fig. 1. Four groups according to operation type: FMD (A-1), FMD+C1L (A-2), FMD+duraplasty (B-1), and FMD+duraplasty+C1L (B-2). LHPF was defined as the area within the anatomy: dorsum sellae, torcular region, and foramen magnum. FMD, foramen magnum decompression; C1L, C1 laminectomy; LHPF, lower half of the posterior fossa.

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went C1L. The magnitude of the operation increased from groups A-1 to B-2 (Fig. 1). There were no exact criteria for selecting surgical techniques; however, larger surgery was performed when severe tonsil herniation or large syrinx volume was noted.

During FMD, the linear skin incision ranged from the inion to the C2 level. The posterior fossa bone was removed at a height of 2.5 cm from the foramen magnum and extended as laterally as possible. At the foramen magnum level, the bone was removed laterally until the internal venous sinus was exposed to ensure that decompression was maximized laterally. Suboccipital ligament splitting was performed using 15 blades without dural opening (A-1). During C1L, the lamina was removed at the same width as the foramen magnum (A-2). The dura layer opened with a large T-shape in the exposed suboccipital area. During dural opening, the arachnoid membrane was also removed to confirm obex opening and CSF circulation. C1L was considered at first, and tonsillectomy was performed in cases of large tonsil herniation, which was inferior to the C1 lower margin. A dural incision was made downward toward the suboccipital ligament without C1L (B-1); however, it expanded up to the lower margin of the C1L (B-2). Allogenic material (Surgisis®; Cook Surgical, Bloomington, IN, USA) was used to repair the dural opening, which was closed using polypropylene sutures (Prolene®; Ethicon, Somerville, NJ, USA) , human fibrinogen (TachoSil®; Baxter, West Hollywood, CA, USA), and fibrin glue (Tisseel®; Baxter).

Postoperative changes in the posterior fossa area and syrinx volume were verified using magnetic resonance imaging (MRI). The first MRI scan was conducted within a week of the operation, and the second one was performed 6 months later. Depending on the degree to which symptoms had improved, a third MRI scan was conducted 1–2 years later to reevaluate changes in the syrinx.

To evaluate the posterior fossa volume, the lower half of the posterior fossa (LHPF) was used within the anatomy of the dorsum sellae, torcular region, and foramen magnum. The LHPF included all of the decompression areas and conveniently allowed the marking of an area on the MRI sagittal image (Fig. 1). FMD in CM-1 is significantly related with caudal posterior fossa, which can be defined as inferior to a line between the dorsum sellae and torcular.[28]

The three-dimensional visualization software AVIEW (version 1.0; Coreline Soft, Seoul, South Korea) was used to measure the syrinx volume. Highly signaled syrinxes within the spinal cord were separated from the CSF. The syrinx areas were calculated in each sagittal cut image, and the volume was calculated as the sum of the areas from each image. Axial reconstruction helped increase volume accuracy (Fig. 2). Using serial pre- and postoperative MRI, syrinx volumes were measured and change slopes determined. The rate of syrinx-volume decrease was analyzed using a statistically linear mixed model including intercept and slope as random effects with SAS version 9.4 (SAS Institute, Inc, Cary, NC, USA).

Unfortunately, syrinx volume was analyzed in only 38 patients (i.e., 28 in B-1 and 10 in B-2) who had undergone surgery between 2007 and 2017. Syrinx-volume measurement was conducted 2 years earlier, and A-1 and A-2 were excluded since their syrinxes were too small to allow volume measurement. Regarding the change in syrinx volume, the effect of C1L was analyzed within the duraplasty groups.

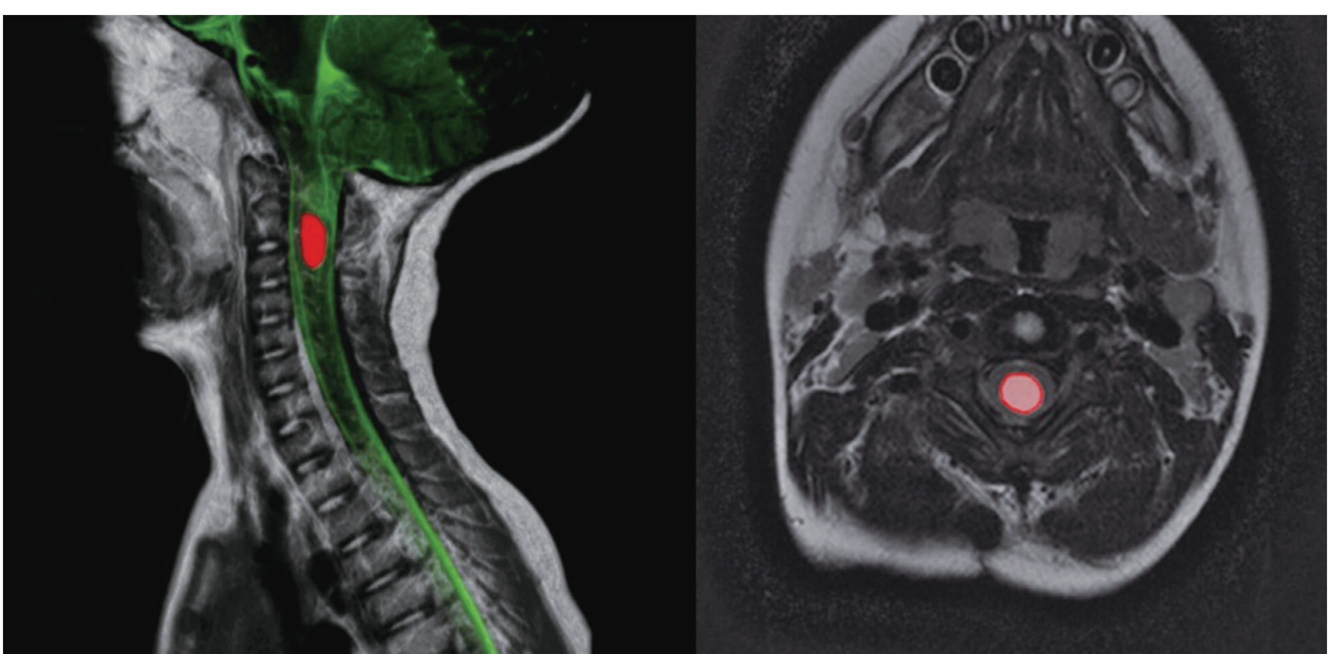

Fig. 2. Sagittal and axial cut of the syrinx on MRI. The CSF area (green) was selected from the original image, and the syrinx area (red) was extracted using AVIEW (version 1.0; Coreline Soft). MRI, magnetic resonance imaging; CSF, cerebrospinal fluid.

Table 2. Differences among the Four Groups

	A-1 (n=6, 6%)	A-2 (n=21, 20%)	B-1 (n=55, 51%)	B-2 (n=25, 23%)	*p* value*
Average tonsil herniation (mm)	11±6	11±6	12±4	14±5	0.027
Postoperative clinical symptoms					
Improved	4 (66)	15 (71)	36 (65)	17 (68)	
Stable	2 (33)	6 (29)	14 (25)	6 (24)	
Aggravated	0	0	5 (9)	2 (8)	
Syringomyelia	2 (33)	5 (24)	43 (78)	18 (72)	
Improved	2 (100)	5 (100)	37 (86)	16 (89)	
Stable	0	0	3 (7)	0	
Aggravated	0	0	3 (7)	2 (11)	
Posterior fossa volume ratio (post–pre)/pre×100 (%)	20±8	23±8	24±7	29±11	0.029
Complications (CSF leakage, CSF diversion repeat Chiari decompression)	0	0	6 (11)	4 (16)	0.526†

CSF, cerebrospinal fluid.
Data are presented as mean±standard deviation or n (%).
**p* value was analyzed using Jonckheere-Terpstra test; †Comparison between B-1 and B-2.

Table 3. Comparison between Duraplasty and Duraplasty with C1 Laminectomy Groups

	B-3 (n=28)	B-4 (n=10)	*p* value*
Median age (yr)	13±16	10±17	
Male-to-female ratio (n:n)	15:13	3:7	
Clinical sign			
Scoliosis	14 (50)	5 (50)	
Hydrocephalus	1 (4)	0	
Average tonsil herniation (mm)	11±5	13±4	0.253
Preoperative syrinx volume (cc)	3.8±2.9	7.3±5.8	0.020
Postoperative syrinx volume (cc)	0.6±0.8	1.1±1.3	0.269
Follow-up period (months)	27±14	29±20	
Rate of decrease per month (%) (*p*-value for time)	5.9 (*p*<0.0001)	7.5 (*p*=0.0001)	0.040

*Independent sample test.

RESULTS

All 107 patients were divided into four groups based on the surgical technique: A-1 (n=6), A-2 (n=21), B-1 (n=55), and B-2 (n=25). The average tonsil herniations were 11, 11, 12, and 14 mm in groups A-1, A-2, B-1, and B-2, respectively (p=0.027). In terms of postoperative clinical symptoms, no aggravations were observed in A-1 and A-2. Fifty (80%) and 23 (92%) patients exhibited improved or stable symptoms in B-1 and B-2, respectively.

Syringomyelia was observed in 2 (33%), 5 (24%), 43 (40%), and 18 (72%) patients in A-1, A-2, B-1, and B-2, respectively. All patients in A-1 and A-2 groups exhibited improved syringomyelia. Of the patients in B-1 and B-2 groups, 93% and 89% demonstrated improved or stable syringomyelia, respectively. The posterior fossa volume increased by 20% in A-1, 23% in A-2, 24% in B-1, and 29% in B-2 (p=0.029). No postoperative complications developed in either A-1 or A-2. However, complications, such as CSF leakage and CSF diversion, occurred in 6 (11%) patients in B-1 and 4 (16%) patients in B-2, with no statistical differences between the two groups (p=0.526) (Table 2).

Among the 38 patients whose syrinx volumes were measured, 28 underwent FMD with duraplasty (B-3) and 10 underwent FMD with duraplasty and C1L (B-4). Two patients from B-3 (7%) and one patient from B-4 (10%) underwent revision due to CSF leakage. One patient (10%) from B-4 underwent reoperation due to worsening of the syrinx; however, all patients eventually exhibited decreased syrinx volumes.

The median patient age was 13±16 years in B-3 and 10±17 years in B-4. The male-to-female ratios were 15:13 in B-3 and 3:7 in B-4. Fourteen (50%) and 5 (50%) patients had scoliosis in B-3 and B-4, respectively. One patient (4%) in B-3 had preoperative hydrocephalus. Preoperative tonsil herniation was 11±5 mm in B-3 and 13±4 mm in B-4. Preoperative syrinx volume was 3.8±2.9 cc in B-3 and 7.3±5.8 cc in B-4 (Table 3).

Syrinx volume decreased by 5.9% and 7.5% per month in B-3 and B-4, respectively. *P*-values were calculated by estimating the effect of time in each group using a linear mixed model. The independent sample test was used for finding statistical difference between B-3 and B-4 (p=0.040) (Table 3).

DISCUSSION

FMD is used to treat CM-1 by decompressing the inferior aspect of the cerebellum, enlarging the total volume of the posterior fossa, and establishing CSF flow.[29] The establishment of CSF flow could not be directly determined in A-1 and A-2. However, certain studies have revealed that bony decompression alone has a favorable effect in patients with CM-1.[30,31] In this study, only bony decompression (A-1 and A-2) was performed in patients with or without small syrinxes. Although syrinx volume could not be calculated in A-1 and A-2, bony decompression was also effective in improving symptoms, with no syrinx aggravation. C1L was associated with increased posterior fossa expansion, both with and without duraplasty. It is a reasonable complementary option when FMD alone is worrisome and duraplasty seems heavy. Moreover, it maximizes posterior fossa expansion with duraplasty. The volume expansion rate was higher when C1L was conducted with duraplasty, and dura incision was perceived to extend downward toward the C-1 lower margin level. However, an increased CSF leakage rate must be considered, based on a longer dura incision.[32,33]

Some studies have reported a 60%–85% prevalence of spinal syrinxes in patients with CM-1, exhibiting consistency with the 64% obtained in our study.[34-36] Decreasing the syrinx volume is a significant challenge when attempting to improve symptoms by restoring normal CSF flow across the foramen magnum.[37] CSF flow can also be estimated indirectly without duraplasty using ultrasonography; however, incomplete obex opening is another important factor that potentially causes insufficient syrinx-volume decrease and cannot be verified without duraplasty. Guo, et al.[38] and Klekamp, et al.[39] found that the arachnoid membrane over the obex hinders CSF flow between the central canal and fourth ventricle. Despite not being analyzed quantitative CSF flow in this study, several patients in B-1 and B-2 groups demonstrated obex adhesion, and we consistently confirmed CSF flow after adhesiolysis or tonsillectomy when duraplasty was conducted. In addition, one patient in B-4 underwent reoperation due to worsening of syrinx. Postoperative adhesion was noted and the syrinx improved after obex reopening. These findings imply that obex opening is also an important factor for decreasing syrinx volume, and performing bony decompression alone can result in surgical failure. Duraplasty is preferred if complete and rapid syrinx-volume reduction is required. In B-1 and B-2, C1L was also associated with improvements in symptoms and the syrinx. Notwithstanding, postoperative complications also increased. In terms of the syrinx-volume decrease rate, C1L decreased syrinx volume more rapidly and was considered related to a more stable CSF circulation.

The larger the syrinx volume, the more aggressive the surgical method selected. Patients in B-2 had approximately twice the preoperative syrinx volume compared to those in B-1 (p=0.02); however, a faster decrease in syrinx volume occurred in B-2 (p=0.04). Postoperative syrinx volumes were 0.6 cc and 1.1 cc, respectively, which had no statistical difference between the two groups (p=0.269). These small volumes resulted in recovery of spinal cord swelling with symptom improvement. Therefore, when a severely large syrinx volume is detected, performing C1L with duraplasty achieves both obex opening and posterior fossa volume expansion more safely.

As a result, C1L was found to offer additional posterior fossa area expansion in CM-1, and proved more effective when duraplasty was included. In terms of syrinx volume, C1L offers a more rapid decrease rate, allowing smoother CSF flow. Duraplasty potentially results in postoperative CSF leakage, and concurrent C1L has a potential to increase the risk of complications. C1L combined with duraplasty is recommended to maximize posterior fossa volume expansion and the syrinx-volume reduction rate. It also augments the effect of bony decompression.

AUTHOR CONTRIBUTIONS

Conceptualization: Dong-Seok Kim. Data curation: Jun Kyu Hwang. Formal analysis: Jun Kyu Hwang. Funding acquisition: Dong-Seok Kim. Investigation: Dong-Seok Kim. Methodology: Kyu-Won Shim. Project administration: Eun Kyung Park. Resources: Kyu-Won Shim. Software: Jun Kyu Hwang. Supervision: Dong-Seok Kim. Validation: Dong-Seok Kim. Visualization: Kyu-Won Shim. Writing—original draft: Dong-Seok Kim. Writing—review & editing: Dong-Seok Kim. Approval of final manuscript: all authors.

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