Sequential Enlargement of Posterior Fossa After Duraplasty for Chiari Malformation Type 1

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■ OBJECTIVE: **There are several material options for duraplasty in surgery for foramen magnum decompression (FMD). We retrospectively analyzed surgical results and the impact of sequential alteration of posterior fossa (PF) size in patients with Chiari malformation type 1 after duraplasty using local fascia.**

■ METHODS: **The patients with Chiari malformation type 1 who underwent FMD with duraplasty using local fascia at our institution between 2004 and 2015 were included in the study. Some pediatric patients who underwent FMD without duraplasty and patients with insufficient follow-up data were excluded. Improvement of clinical signs, regression of syrinx, and temporal alteration of PF size were analyzed with perioperative magnetic resonance imaging. Postoperative clinical outcomes were evaluated at the final follow-up using the Chicago Chiari Outcome Scale. Correlations between the PF size alteration and size of dural patch or Chicago Chiari Outcome Scale were statistically analyzed.**

■ RESULTS: **Twelve patients were included in this study. The mean age of the patients was 31.9 (range, 18–48) years. Syrinx was incorporated in 11 patients and regressed postoperatively in all patients. The PF was significantly enlarged sequentially over time ($P < 0.05$). The degree of enlargement was positively correlated with size of the fascial patch ($r = 0.540$). Postoperative clinical outcomes at final follow-up were positively correlated with the degree of PF enlargement ($r = 0.678$).**

■ CONCLUSIONS: **The PF was enlarged sequentially over time after duraplasty using local fascia. The degree of enlargement was positively correlated with the size of the fascial patch and the likelihood of a favorable postoperative outcome.**

INTRODUCTION

It is well known that patients with Chiari malformation type 1 (CM1) have a small or overcrowded posterior fossa (PF), and reduced height of the PF is the main cause in most patients.[1-3] Treatment for patients with CM1 aims to decompress neural structures and restore normal cerebrospinal fluid (CSF) dynamics at the cranio-vertebral junction. This treatment approach is usually successful in achieving regression of associated syringomyelia. The gold standard treatment for CM1 is foramen magnum decompression (FMD).[4] However, there is no general agreement on how this decompression should be performed.[5,6] There are several proposed techniques for FMD, ranging from extradural decompression alone to intra-arachnoid tonsillar resection. Even in the most standard extra-arachnoidal approach, which involves suboccipital craniectomy with duraplasty, there are several material options for dural patch in duraplasty.[4,7-10] Although numerous reports have compared surgical techniques for duraplasty,[4,5,9] none has clearly identified a nonsurgical factor likely to maximize the likelihood of a successful clinical outcome.

Key words
- Chiari malformation
- Duraplasty
- Foramen magnum decompression
- Local fascia
- Posterior fossa

Abbreviations and Acronyms
CM1: Chiari malformation type 1
CSF: Cerebrospinal fluid
FMD: Foramen magnum decompression
MRI: Magnetic resonance imaging
PF: Posterior fossa
SD: Standard deviation

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Factors previously identified as being negatively associated with successful postoperative outcomes in patients with CM1 include symptomatic central cord pathology and syringomyelia,[11-14] bulbar symptoms and ataxia,[15] and duration of symptoms.[16] Normal preoperative hindbrain CSF flow on cine-magnetic resonance imaging (MRI)[17] has also been proposed as a factor negatively associated with successful postoperative outcomes. Several reports have proposed a relationship between MRI-based PF volume and outcomes after FMD.[18,19] However, no report has assessed postoperative temporal alteration in PF size and its relationship with clinical outcomes after FMD for CM1. Therefore, we retrospectively analyzed surgical results at our institute and the impact of sequential change of PF size with time in patients with CM1 who underwent FMD with duraplasty using local fascia.

METHODS

All experiments followed the tenets of the Declaration of Helsinki. All research protocols were approved by the institutional review board of Nara Medical University, and the need for informed consent was waived. (approval number: 1654).

Study Design

We retrospectively studied all charts of patients with CM1 who underwent FMD at our department in Nara Medical University Hospital between January 2004 and December 2015. Although FMD was done in 22 patients in this period, 7 pediatric patients who underwent FMD with dural dissection of the outer layer only during this period were excluded. Three patients who had insufficient follow-up clinical and radiologic data were also excluded, 12 patients who underwent FMD with duraplasty using local fascia were finally included in this study.

Surgical Technique

The patient was placed in prone position with the head in flexion. A midline skin incision was made from the inion to the spinous process of the axis. Local fascia of the trapezius muscle was collected first, followed by suboccipital craniectomy with FMD and C1 laminectomy (Figure 1A and B). The size of corrected fascia varied considerably in each case. The diameter of the cranial opening was defined by the individual transverse diameter of the foramen magnum, and approximately 30 x 30 mm in all cases. Based on the position of the cerebellar tonsils, the posterior arch of axis was occasionally partially drilled out. After cutting the thickened cranio-cervical dural band, dural incision, which is Y-shaped in normal cases, was made while taking care not to tear the arachnoid membrane. If the arachnoid tear was apparent, arachnoid repair was performed using absorbable hemostat agent with fibrin glue. To achieve watertight closure, expansive duraplasty was subsequently performed with the collected fascia (Figure 1C).

Clinical Data Assessment

Our data were retrospectively gathered from medical records, radiographic images, intraoperative photos, and pre- and postoperative neurologic examinations. We analyzed clinical and radiologic characteristics and outcomes. Particularly, improvement of clinical signs, regression of syrinx, and temporal alteration of PF size with perioperative MRI was analyzed. Postoperative clinical outcome at final follow-up was estimated using the Chicago Chiari Outcome Scale (CCOS).[20,21]

Perioperative PF size was calculated from the middle section of sagittal T2-weighted imaging obtained with MRI, at 3 points in time: preoperatively, 2 months postoperatively, and >1 year postoperatively. The perioperative PF size was estimated using the invariable anatomic lines: between the posterior lower edge of the C2 vertebral body to the lower edge of the C2 spinous process and the posterior clinoid process to the cranial edge of the tentorium cerebellum (Figure 2A). The measurements of PF size were performed using a workstation (SYNAPSE VINCENT, Fujifilm, Tokyo, Japan) by a neurosurgeon who is independent from interpretation of the study. The individual size of dural defect was

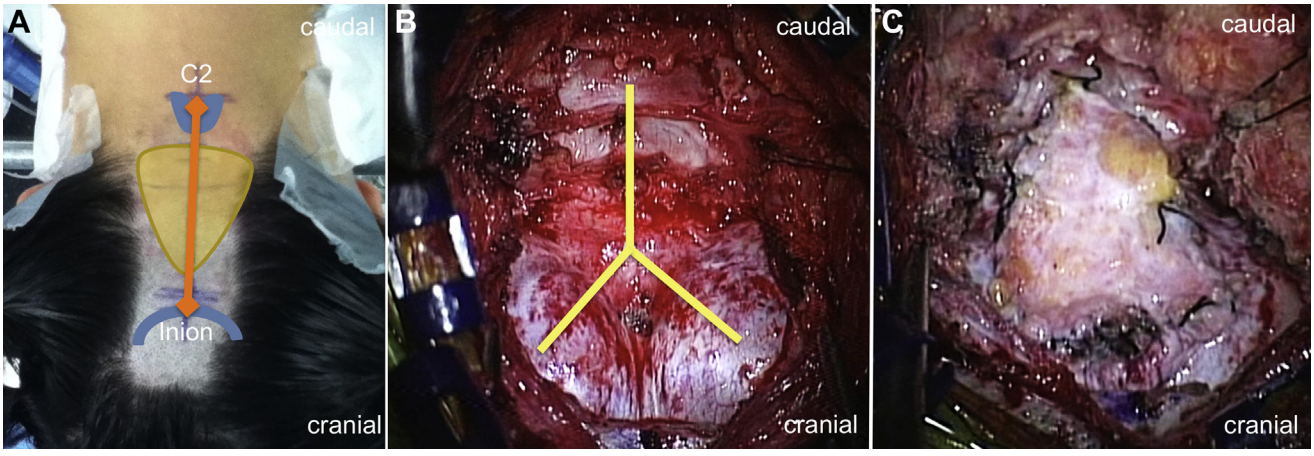

Figure 1. Surgical techniques of illustrative case. (**A**) A skin incision was made from the inion to the spinous process of the axis, and local fascia was collected from the trapezius muscle. (**B**) Suboccipital craniectomy with foramen magnum decompression and C1 laminectomy was performed, followed by dural incision. Y-shaped incision was made in normal cases. (**C**) Expansive duraplasty using the collected fascia was performed.

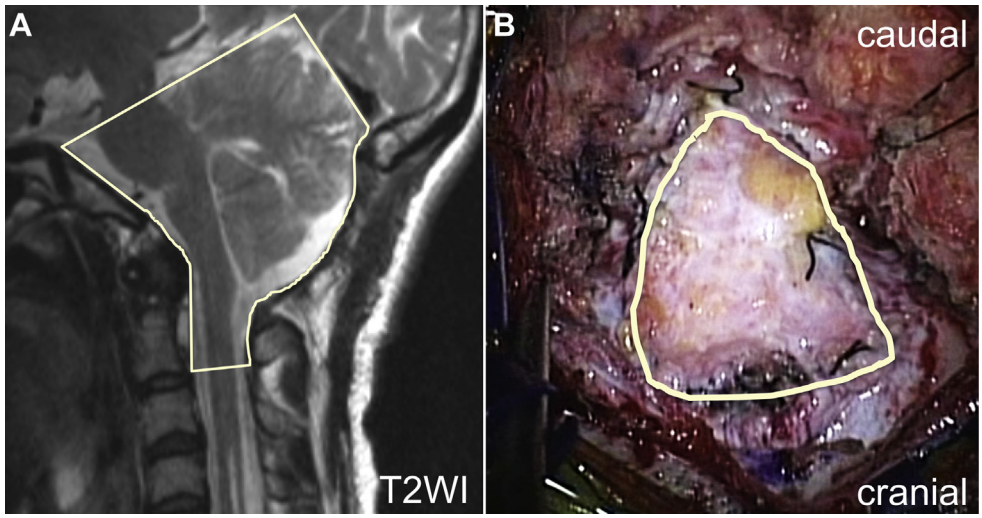

Figure 2. Clinical data assessments. (**A**) Assessment of posterior fossa size. The perioperative posterior fossa size was estimated by T2-weighted image of sagittal magnetic resonance imaging using the invariable anatomic lines; between posterior lower edge of C2 vertebral body to lower edge of C2 spinous process, and posterior clinoid process to cranial edge of tentorium cerebellum (*enclosed line*). (**B**) Assessment of the area of dural defect. The size of dural defect was calculated by intraoperative microscopic findings (*enclosed line*). T2WI, T2-weighted image.

also measured just after duraplasty by intraoperative microscopic findings (Figure 2B). For a given dural defect size, any number of patch sizes could be envisioned.

Postoperative sequential changes in the size of the PF were compared across time points. The relationship between the size of the dural defect and degree of PF enlargement (from preoperative assessment to final follow-up) was investigated. Moreover, relationship between the CCOS score and degree of PF enlargement from preoperative assessment to final follow-up was also assessed.

Statistical Analysis

All statistical comparisons were performed using PASW Statistics software version 18 (IBM SPSS, Armonk, New York, USA). Postoperative sequential changes in the size of the PF with time were compared to the Wilcoxon signed-rank test. The relationship between the size of the dural defect and degree of PF enlargement was assessed using the Pearson's product moment correlation coefficient. The relationship between the CCOS score and degree of PF enlargement was assessed with the Spearman's rank correlation coefficient. Values are presented as mean ± standard deviation (SD). Differences were considered significant at $P < 0.05$.

RESULTS

Clinical Characteristics

Twelve patients underwent surgical decompression for CM1 during the study period. Detailed characteristics of the 12 patients with CM1 are presented in Table 1. The study population included 3 men and 9 women aged 18–48 years [mean ± SD, 31.9 ± 12.3 years]. Symptoms included occipitalgia on strain in 7 cases (58.3%), subjective sensory disturbance in 7 cases (58.3%), and dissociative sensory deficit in 8 cases (66.7%). MRI revealed syrinx formation in 11 patients (91.7%). Mean distance of the cerebellar tonsil herniation below the foramen magnum ranged from 5.1–14.6 mm (mean ± SD, 10.3 ± 2.8). The duration of postoperative follow-up ranged from 12–68 months (mean ± SD, 31.6 ± 24.3) (Table 1).

Postoperative Improvement at Final Follow-Up

Postoperative regression of syrinx was observed in all patients. Headache on strain, subjective sensory disturbance, and dissociative sensory deficit were improved postoperatively in 100%, 71%, and 0% of patients, respectively (Table 2).

Only 1 minor surgical complication was noted. One patient experienced subcutaneous CSF leakage, which was spontaneously resolved without additional treatment.

Sequential Changes in Size of the PF with Time

Statistical analysis of sequential changes in the size of the PF revealed a significant difference over time ($P < 0.05$) (Figure 3). Use of a local fascial patch appeared to enhance PF enlargement at the final follow-up.

Relationship Between the Size of Dural Defect and Degree of PF Enlargement

In the statistical analysis of the relationship between the size of dural defect and degree of PF enlargement measured on sagittal MRI, the degree of PF enlargement was positively correlated with the size of fascial patch ($r = 0.540$) (Figure 4). Use of a larger fascial patch was correlated with a greater increase in the postoperative size of the PF.

Table 1. Clinical Characteristics of the Cases

Variables	No.
Number of Cases (male/female)	12 (3/9)
With syrinx	11
Mean age (years)	31.9 (18—48 years)
The mean distant of cerebellar tonsil herniation below foramen magnum	10.3 mm (5.1—14.6 mm)
Follow-up periods	31.6 ± 24.3 months

Relationship Between the Clinical Outcome and Degree of PF Enlargement

In the statistical analysis of the relationship between the CCOS score and degree of PF enlargement, postoperative clinical outcome at the final follow-up had a positive correlation to the degree of PF enlargement (r_s = 0.678) (Figure 5). A greater degree of PF enlargement was associated with improved clinical outcomes.

ILLUSTRATIVE CASE

An 18-year-old woman suffered from headache at straining and numbness in both hands. Neurologic examination revealed dissociative sensory disturbance below the C6 spinal cord segment. Cervical MRI revealed 10.9 mm cerebellar tonsil herniation into the foramen magnum with cervical syringomyelia (Figure 6A). She was diagnosed as symptomatic CM1 with syringomyelia and underwent FMD with duraplasty using local fascia. MRI at 2 months after the surgery revealed expansion of syrinx, despite enlargement of PF (Figure 6B). On the other hand, postoperative clinical course was uneventful and her headache at straining and hand numbness resolved, except for dissociative sensory disturbance, therefore she was taken conservative follow-up. Thirty-eight months later, MRI revealed resolution of the cerebellar tonsil herniation and the regression of the cervical syringomyelia, with additional PF enlargement (Figure 6C).

DISCUSSION

This study presents data demonstrating a correlation between the degree of postoperative PF enlargement and size of the dural defect for duraplasty as well as clinical outcomes in patients who underwent FMD with localized fascia. Moreover, significant sequential enlargement of the PF with the passage of time was observed postoperatively.

Table 2. Clinical Features and Outcomes of the Cases at Final Follow-Up

Clinical Features	Improvement Ratio
Regression of syrinx	100% (11/11 cases)
Headache on strain	100% (7/7 cases)
Subjective sensory disturbance	71% (5/7 cases)
Dissociative sensory disturbance	0% (0/8 cases)

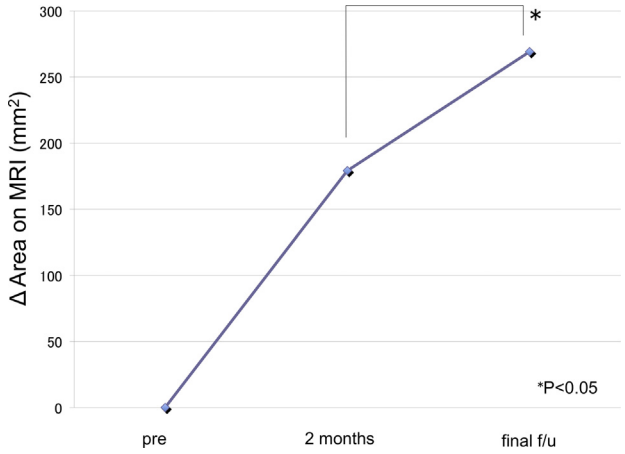

Figure 3. Postoperative sequential changes in size of the posterior fossa. Posterior fossa volume was enlarged sequentially with the passage of time with significant difference ($P < 0.05$). f/u, follow-up; MRI, magnetic resonance imaging.

Surgical Variation and Postoperative Clinical Outcomes of FMD for CM1

The main cause of CM1 is a small or overcrowded PF,[1-3] FMD remains the first-line treatment for CM1. However, there are several proposed techniques for FMD in patients with CM1. These include partial suboccipital craniectomy only with dural dissection of the outer layer, suboccipital decompression with duraplasty, and suboccipital decompression with duraplasty and arachnoid dissection.[7] These 3 techniques could be considered as extradural, extra-arachnoidal, and intradural approaches, respectively. There

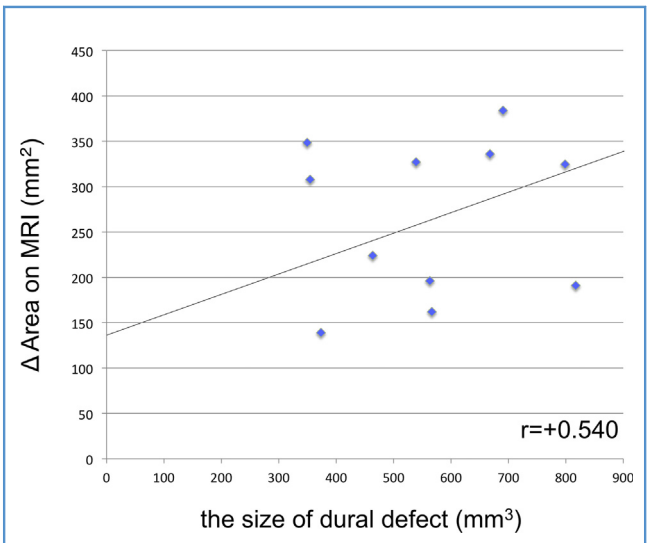

Figure 4. Relationship between the size of dural defect and the posterior fossa enlargement. The degree of posterior fossa enlargement was positively correlated with the size of fascial patch (r = 0.540). MRI, magnetic resonance imaging.

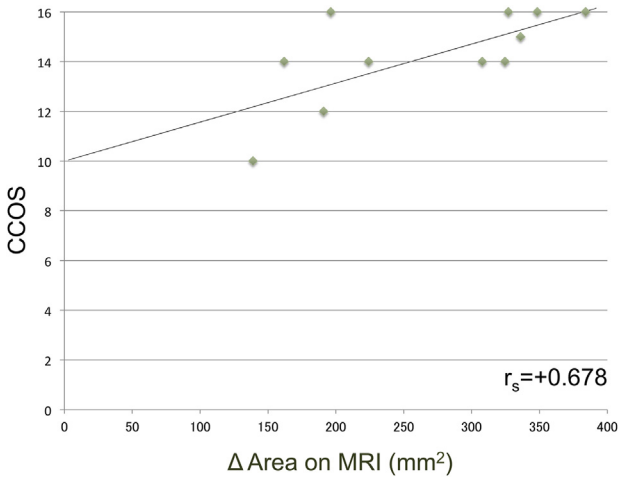

Figure 5. Relationship between the clinical outcome and the degree of posterior fossa enlargement. Postoperative clinical outcome at final follow-up had positive correlation to the degree of posterior fossa enlargement. (r_s = 0.678) CCOS, Chicago Chiari Outcome Scale; MRI, magnetic resonance imaging.

are some reports regarding the surgical results and complication among these techniques.[5,22] The intradural technique carries an increased risk for complications like wound infection, CSF leakage, pseudomeningocele, and meningitis. The extradural technique is associated with a lower rate of complications than FMD with duraplasty. However, the rate of clinical response when using the extradural technique is also lower than when using FMD with duraplasty.[5]

Experts agree that the optimal approach for FMD requires the improvement of CSF flow through the foramen magnum to avoid postoperative arachnoiditis. To keep the arachnoid plane intact, we chose the extra-arachnoidal technique for our surgical approach. Sindou and Gimbert[7] also suggested that extra-arachnoidal technique with preservation of the arachnoid membrane had favorable results. Even if postoperative radiologic improvement is achieved, it is another issue that clinical improvement would be obtained. From the perspective of the relief of clinical impairment, the discussion has been unsatisfactory. Although relief of clinical signs following decompression has been reported in up to 75%–90% of patients with CM1,[23-25] as many as 35%–45% may have minimal to no relief years after surgery.[14,16,26] Hence, identification of novel predictors of better outcomes are needed to select patients most likely to benefit from surgical intervention. We therefore planned this study to identify factors related to successful postoperative outcomes after use of the extra-arachnoidal technique for patients with CM1.

FMD for Patients with Developed Occipital Sinus

We occasionally encounter a case of CM1 with the development of occipital sinus, which represents a barrier to dural incision because of effects on venous circulation from the brain. In such situations, it may be possible to create a unilateral curvilinear dural incision before performing duraplasty.[27] These study results would be applicable in such a severe clinical situation. The advantage of the presented technique in such a situation is that even a small dural opening will take a bigger PF enlargement sequentially after surgery.

There are also several options for materials used for duraplasty in FMD for patients with CM1,[7,8] without reaching a consensus about the best suited one. Some of the choices for the materials include autogenous grafts and artificial dura.[4] We speculated that creation of a larger dural opening without arachnoid manipulation would show superior postoperative results. These positive outcomes could be enhanced using local fascia as the duraplasty material. Not only local fascia but also other autogenous materials, such as local periosteum, may be similarly used. Moreover, under selection of artificial dura, not only a larger dural incision but also a larger artificial dural patch may lead to better postoperative outcomes.

Novel Factors Regarding Better Surgical Outcomes of FMD

There appears to be a relationship between radiologic assessment of PF volume and clinical outcomes. Badie et al.[28] reported that patients with a small PF ratio, which is the ratio of the PF volume to supratentorial volume, tend to respond better to suboccipital decompression. Moreover, there are several studies reporting that the degree of postoperative PF enlargement contributes to postoperative clinical outcomes,[19] and the present study results comply with the same.

Regarding the extra-arachnoidal technique in FMD for patients with CM1, there are few detailed reports about the surgical maneuvers necessary to achieve superior clinical outcomes. Although there are some choices for the materials for duraplasty in FMD,[4] using local fascia of trapezius muscle, located at the surface of posterior cervical muscles, has an advantage of requiring a shorter skin incision than when using periosteum located at the parietal bone. Moreover, cervical fascia is an elastic-like material allowing for a stronger watertight closure[4] rather than artificial dura.

This illustrative case may show other possibilities of this surgical technique using local fascia. The case particularly followed the impressive course on postoperative radiologic examinations. Although PF is enlarged postoperatively, CSF flow retention through foramen magnum unfortunately gives rise to syrinx enlargement that may occur owing to surgical manipulation-induced local environmental change (e.g., cerebellar tonsil repositioning or adhesive arachnoiditis). Because there was no manipulation to the arachnoid membrane intraoperatively and postoperative clinical course was uneventful, conservative follow-up was performed without any additional procedure. Although it was unknown why syrinx was significantly turned to regression although the conservative follow-up, it was speculated that postoperative sequential enlargement of PF due to duraplasty using local fascia positively contributed to this amazing phenomenon.

In the present study, it was first observed that PF enlarges sequentially with time after FMD with local fascia. The result of this study demonstrated that the use of local fascia has a substantial additional effect to facilitate PF enlargement, which directly contributes to postoperative better outcomes.

Limitations

There are some major limitations of the current study, largely arising from the retrospective design and its small sample size. Another limitation is that there was no control group in this study.

Figure 6. Cervical magnetic resonance imaging (MRI) of the illustrative case. Sagittal MRI of the illustrative case at the 3 points in time revealed gradual enlargement of posterior fossa (PF) size, and transient enlargement of the syrinx. Preoperative MRI (**A**) and MRI at 2 months after the surgery (**B**) reveal expansion of syrinx, despite of enlargement of PF. At final follow-up, PF enlarges additionally, and the syrinx achieved a complete regression spontaneously without any additional treatment (**C**). T2WI, T2-weighted image.

The possibility of gradual increase in PF size with suboccipital craniectomy alone could not be denied completely. Moreover, PF enlargement was estimated by an MRI-based measurement, which measures changes in a 2-dimensional area. Therefore, prospective data analysis using a 3-dimensional analysis is needed to confirm the results presented earlier.

CONCLUSIONS

We retrospectively analyzed surgical results and temporal alteration of PF size in patients with CM1 who underwent FMD with local fascial patch without arachnoid manifestation. In such patients, the PF enlarges sequentially with time and the degree of enlargement is positively correlated with the size of dural defect. Moreover, the size of dural defect and postoperative clinical outcome positively correlates with postoperative PF enlargement.

Therefore, a larger dural opening would depend on greater postoperative results, and additional impact with local fascia as a duraplasty material would be expected. These results also provide more helpful information especially about patients with developed occipital sinus in whom dural incision is particularly limited.

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