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Comparison between Dura-Splitting Technique with Duraplasty in Symptomatic Patients with Chiari Malformation Type I: A Systematic Review and Meta-analysis

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Abstract

BACKGROUND: There are many surgical procedures for CIM patients, posterior fossa decompression with fibrous band excision, with additional duraplasty, or syringosubdural shunt for syringomyelia related CIM. Prospective studies have been carried out but yet no conclusion, on which one is the best option. The objective of this study was to assess qualitatively the outcome of posterior fossa decompression with dura-splitting (PFDDS) technique compared to posterior fossa decompression with duraplasty (PFDDP) for treating CIM patients.

AIM: This study aimed to give us a preference while conducting surgery in a patient with Chiari malformation type I (CIM) between posterior fossa decompression with incision of the fibrous band of the dura (dura-splitting/DS) technique and duraplasty (DP) technique.

METHODS: The analysis conducted using PRISMA flowchart with PICO framework (Patient: Chiari malformation type I patient over preschool age; Intervention: Dura-splitting; Comparison: Duraplasty; and Outcome: Complication rate, length of stay, reoperation rate, syrinx reduction, symptomatic improvement, and operation time) and already registered for meta-analysis study with database searching from PubMed, the Cochrane Library, and Google Scholar that following inclusion criteria: (1) Original study; (2) study that compares DS and DP in CM- I; and (3) patient age over preschool age.

RESULTS: A review of five included studies involving 458 patients met the inclusion criteria, in which 319 patients treated with DS surgery and 139 for DP surgery for this study. Significantly DS technique correlated lower rate of complication (RR = 0.20; p < 0.0001), shorter length of stay (MD = −3.53; p = 0.0002), and shorter operation time (MD = −58.59; p = 0.0004). No significant differences in reoperation rate (RR = 1.90; p = 0.22), symptom improvement (RR = 1.12; p = 0.44), and syrinx reduction (RR = 1.11; p = 0.56) were noted.

CONCLUSIONS: Posterior fossa decompression using the DS technique is associated with a lower rate of complication, shorter length of stay, and shorter operation time. However, no significant differences were found in the reoperation rate, symptom improvement, and syringomyelia reduction between these two techniques.

Introduction

Chiari malformation type I (CIM) is one of the Chiari malformation subgroup defined as cerebellar tonsillar herniation below the foramen magnum more than 5 mm [1]. This entity differs from the other as no brain stem nor fourth ventricle involvement [1], [2]. The herniated tonsil may obstruct cerebrospinal fluid flow at the level of occipitocervical junction [3], [4]. Obstruction of the cerebrospinal fluid flow leads to increasing of intracanal pressure then formation of the syrinx. This malformation rarely correlates with other intracranial anomalies that also differ from other subgroup of Chiari malformation.

Chiari malformation type I tends to be a radiological assessment as this founding on neuroimaging does not related to patient symptoms. Only about 7% of these patient will develop symptoms related to Chiari malformation [5]. There are three main etiologies that caused symptoms development in CIM such as cerebellar dysfunction, brainstem problem, and spinal cord dysfunction. These symptoms easier to detected on children, especially they on preschool age and older. Furthermore, as mentioned before the definition of Chiari malformation type 1 malformation, this entity best diagnosed using MRI as this imaging can evaluate cerebellar tonsil, fourth ventricle and brain stem.

Nowadays, several surgical procedures have been emphasized to treat patient with Chiari malformation type I. The mainstay of surgical procedure is to decompress the posterior fossa and releases the pressure on the tonsils that obstruct cerebrospinal fluid flow and lead to syrinx formation and improve patient condition. However, decompressing posterior fossa may be achieved using varied surgical procedures details, including the extension of dura opening, the use of dura-graft material, shunting intrasyrinx, and several medication [1], [6].

Some authors emphasized that simple posterior fossa decompression by suboccipital craniotomy with additional splitting of fibrous band of the dura is preferred due to minimal complication and bleeding. On the other side, performing additional duraplasty either using autograft or allograft may provide promising result as it gives more space to the posterior fossa [6].

Therefore, we conduct this study to compare the patient outcomes between these two surgical procedures in managing patient with Chiari malformation type I. We evaluate the complication rate, length of stay, reoperation rate, syrinx reduction, symptomatic improvement, and operation time.

Methods

Eligibility criteria

Eligibility criteria were made based on our PICO framework. All studies compare posterior fossa decompression with dura-splitting (PFDDS) technique and with duraplasty for treating patient with symptomatic Chiari malformation type I. Articles were restricted to the English and Bahasa only. Both prospective and retrospective cohort studies were included in the study. There is no limitation on publication year. Cadaveric, anatomical, animal studies, review article, and qualitative studies were excluded from the study.

Chiari malformation type I was defined as any cerebellar tonsillar herniation below the foramen magnum projecting to the spinal canal. We included that both sexes and age restriction were applied for patient over preschool age.

PFDDS technique defined as suboccipital craniotomy exposing posterior fossa dura with splitting the fibrous band over it as leave the inner duramater intact. Posterior fossa decompression with duraplasty (PFDDP) technique defined as suboccipital craniotomy exposing posterior fossa dura with incision of the dura and performing duraplasty either using allograft or autograft. Any articles that did not mention the surgical technique were excluded from the study.

The outcome of this study was complication rate, length of stay, reoperation rate, syrinx reduction, symptomatic improvement, and operation time. Minimal evaluation of the outcome is strict to 1 year.

Literature search strategy

Literature search was conducted based on preferred reporting items for systematic review and meta-analysis (PRISMA) guidelines. We conduct literature search using Cochrane, Google Scholar, and PubMed using Boolean operator of combination on these terms: "Chiari malformation type I," "dura-splitting," "duraplasty," and "outcome." We restrict an English language study only. Manual searching was conducted by listing all the reference list from all eligible articles.

Data collection

All studies were evaluated and selected by two authors TJ and ST to minimize selection error. Evaluated data that collected from each study were: (1) Patient characteristics, (2) surgical approach, and (3) outcomes. If any contradictory selection occurred, the decision was discussed with other authors to have the conclusion.

Assessment of quality of study

Studies that pass the selection criteria were assessed to ensure the research validity. Quality assessments were conducted using Newcastle-Ottawa Scale (NOS) as it already standardized and minimize the bias possibility. The cutoff point for final decision of the quality of study were six (with nine as total point for NOS). Studies that scored over the cutoff point classified as high-quality studies and otherwise will be judged.

Statistical analysis

All data were collected and analyzed using Review Manager Software (version 5.4). Pooled data were then grouped using random-effects or fixed-effects based on the heterogeneity between all studies.

Results

Studies characteristics

Based on our database searching, a total of 439 were found with three studies identified from manual searching. We included five studies that met our criteria after undergo systematic searching on database (Figure 1). All of the studies were observational studies. A total of 460 patients were included with 319 of patients underwent PFDDS and the rest underwent PFDDP. Based on NOS scoring, these five studies considered as high-quality studies. Characteristics are listed below in Table 1.

Quantitative analysis

Complication rate

All five studies with total of 319 patients underwent PFDDS and 141 patients underwent

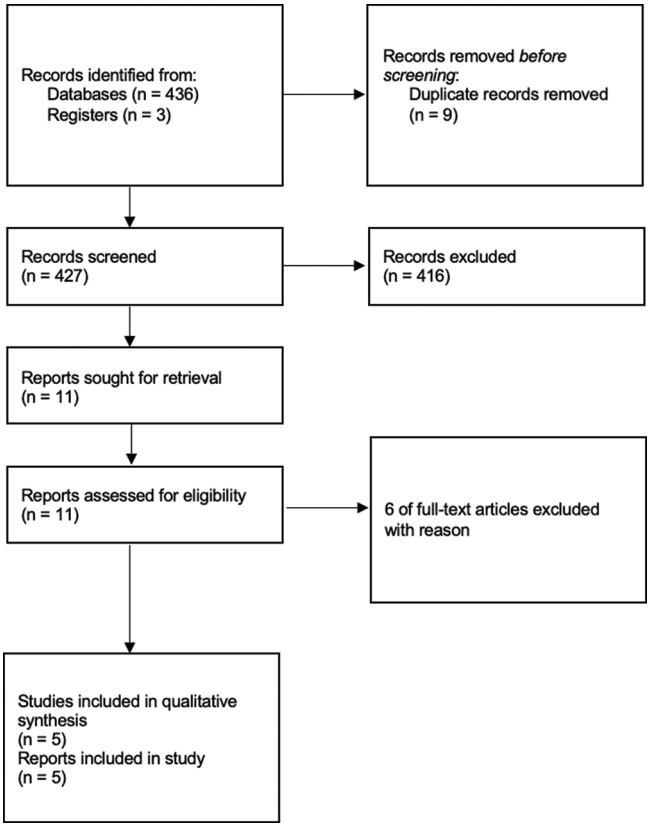

Figure 1: Flowchart of identification of the study

PFDDP reported the complication rate of the surgery (Figure 2). Complication were include aseptic meningitis, pesudomeningocele, wound infection, and CSF leaks. We were using fixed effect model, as the heterogeneity among these five studies was low (I^2 = 0%; p = 0.85). The meta-analysis indicates that there is significance difference in decreasing complication rate using PFDDP technique with RR of 0.20 (p < 0.0001; 95% CI, 0.09–0.44).

Length of stay

Two studies, with total of 272 patients, were included with total of 201 patients underwent PFDDS and 71 patients underwent PFDDP reported the length of stay (Figure 3). We were using fixed effect model, as the heterogeneity was low (I^2 = 0%; p = 0.49). The meta-analysis indicates that there is significance reduction in length of stay using PFDDP technique with MD of −3.53 (p = 0.0002; CI, [−5.40]–[−1.66]).

Reoperation rate

Three studies, with 118 patients underwent PFDDS and 70 patients underwent PFDDP, reported the reoperation rate (Figure 4). We were using fixed effect model as the heterogeneity was low (I^2 = 48%; p = 0.15). The meta-analysis indicates that there is not significance different between these two groups with RR of 1.90 (p = 0.22; CI, 0.69–5.23).

Operation time

A total of two studies, with 201 patients underwent PFDDS and 71 patients underwent PFDDP, reported the operation time (Figure 5). We were using random effect model, as the heterogeneity was high (I^2 = 70%; p = 0.07). The meta-analysis indicates that there is significance reduction in operation time using PFDDP technique with MD of −58.59 (p = 0.0004; CI, [−56.92]–[−39.31]).

Syringomyelia reduction

A total of three studies, with 199 patients underwent PFDDS and 57 patients underwent PFDDP, reported the rate of syringomyelia and its reduction (Figure 6). We were using fixed effect model, as the heterogeneity was high (I^2 = 56%; p = 0.10). The meta-analysis indicates that there is not significance different between these two groups with RR of 1.11 (p = 0.56; CI, 0.79–1.56).

Symptoms improvement

A total of four studies, with 241 patients underwent PFDDS and 106 patients underwent PFDDP reported the

Table 1: Characteristic of the studies

Study Author	Type of study	Gender		Intervention	Control	Outcome
Ilyas, 2000	Retrospective Observational	Male: 4 Female: 7	Male: 7 Female: 16	Dura Splitting 11 patients	Duraplasty 23 patients	- Syrinx reduction (DS: 4; DP: 4) - Reoperation rate (DS: 2; DP: 0) - Symptoms improvement (DS: 8; DP: 20) - Complication (DS: 1; DP: 10)
Chotai, 2014	Retrospective Observational	n/a	n/a	Dura-splitting 29 patients	Duraplasty 12 patients	- Reoperation rate (DS: 0; DP: 1) - Symptoms improvement (DS: 28; 7) - Complication (DS: 1; DP 4)
Geng, 2018	Retrospective Observational	Male: 6 Female 11	Male: 7 Female: 25	Dura-splitting 17 patients	Duraplasty 32 patients	- Length of Stay (DS: 10 days; DP 12.61 days) - Operation Time (DS: 167.94 mins; DP 248.03 mins) - Symptoms improvement (DS: 13; DP: 26) - Complication (DS: 0; DP: 10)
Oral, 2018	Retrospective Observational	Male: 22 Female: 56	Male: 21 Female: 14	Dura-splitting 78 patients	Duraplasty 35 patients	- Syrinx reduction (DS: 7; DP: 3) - Reoperation rate (DS: 10; DP 2) - Complication (DS: 4; DP: 8)
Sajan, 2020	Retrospective Observational	Male: 41 Female: 143	Male: 13 Female: 26	Dura-splitting 184 patients	Duraplasty 39 patients	- Syrinx reduction (DS: 21; DP: 11) - Length of Stay (DS: 14 days; DP: 18 days) - Operation Time (DS 117 mins; DP: 163 mins) - Symptoms improvement (DS: 124; DP: 20) - Complication (DS: 2; DP: 1)

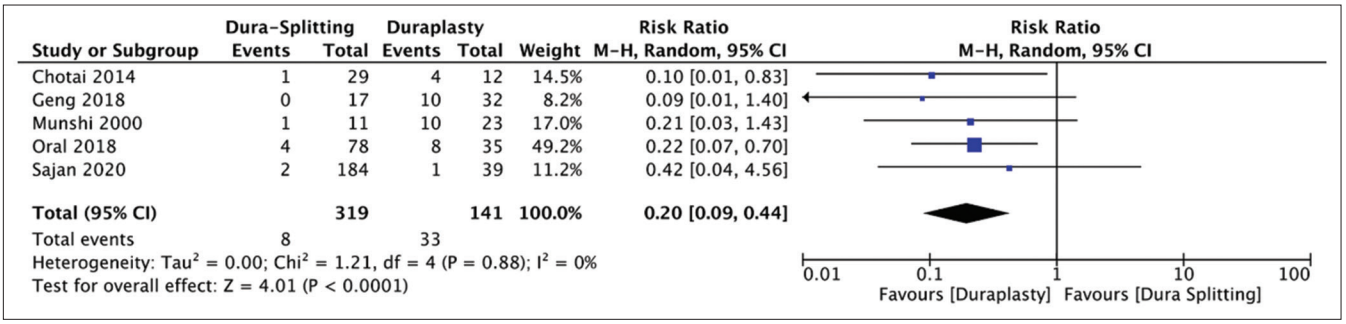

Figure 2: Complication rate

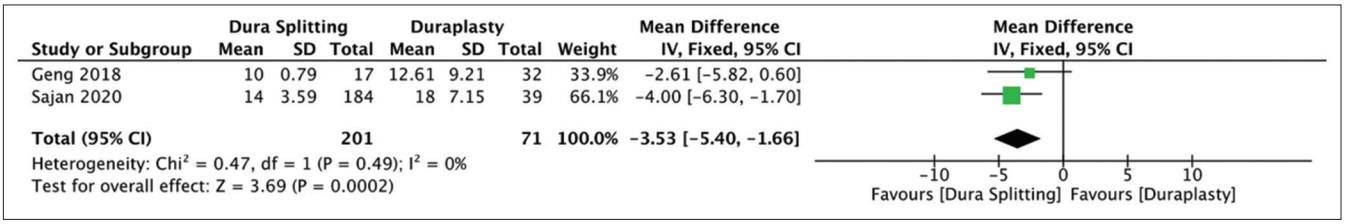

Figure 3: Length of stay

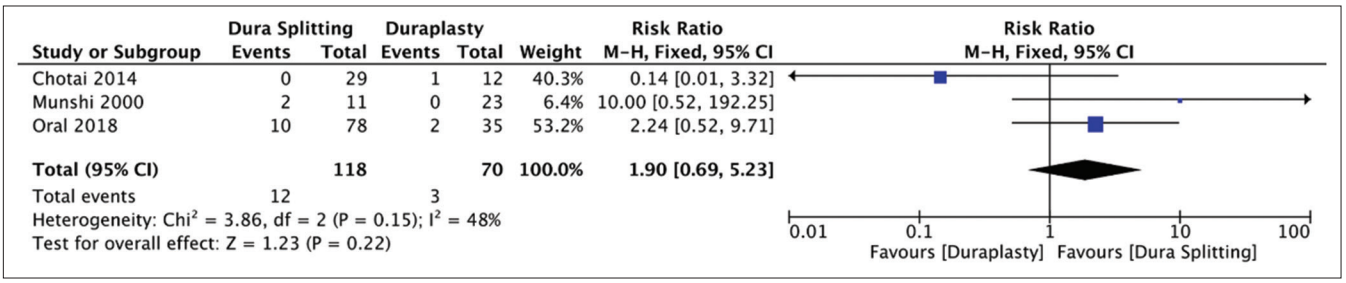

Figure 4: Reoperation rate

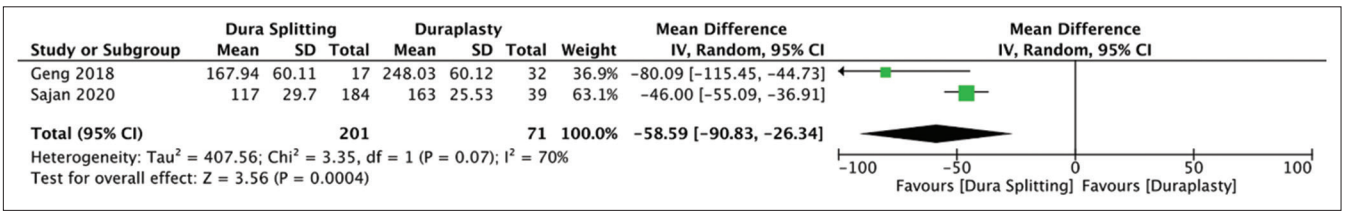

Figure 5: Operation time

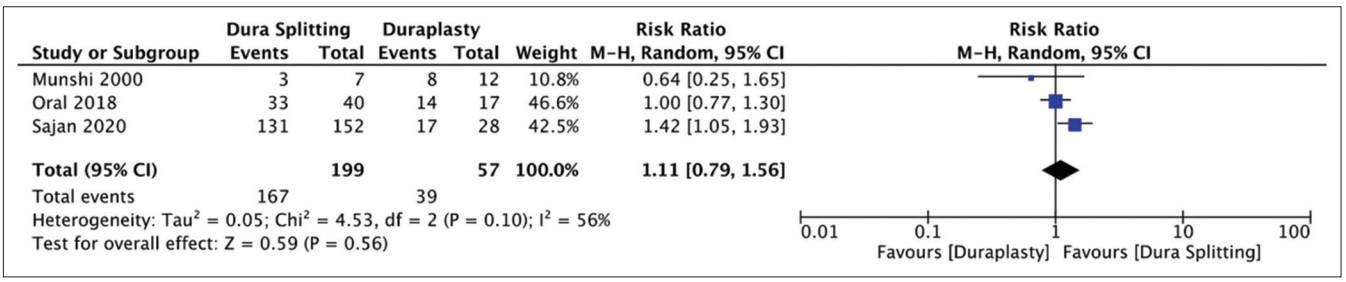

Figure 6: Syringomyelia reduction

rate of syringomyelia and its reduction (Figure 7). We were using random effect model, as the heterogeneity was high (I^2 = 59%; p = 0.06). The meta-analysis indicates that there is not significance different between these two groups with RR of 1.12 (p = 0.44; CI, 0.84–1.50).

Discussion

Chiari malformation type I (CIM) is one of Chiari malformation subgroup classification. There

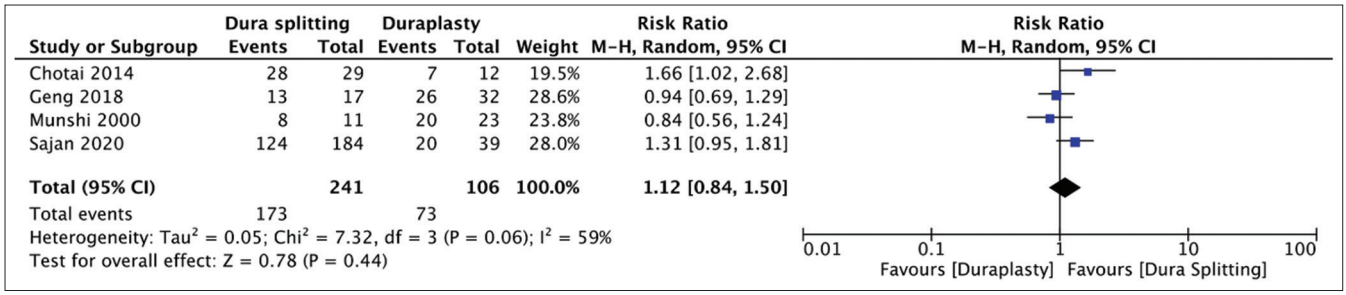

Figure 7: Symptoms improvement

are many problems which occur caused by tonsillar herniation, either due to CSF flow abnormality, brainstem, or cerebellar compression. These symptoms also ranging from mild to severe problem of the patients [7], [8], [9]. Symptoms of the patient are fluctuate that follow the dynamic intracranial pressure and may be induced by Valsalva [2]. Younger children tend to present with cranial nerve problem as it difficults to evaluate their symptoms too. Spinal cord may be compressed either by the herniated cerebellar tonsil or syrinx formation [10]. Although debates regarding the pathophysiology of syringomyelia in patient with CIM is still ongoing, there is one agreement the importance of posterior fossa decompression to treat CIM with or without syringomyelia [11].

Syrinx formation may be related to imbalance of CSF flow between cranial and spinal compartment due to brainstem herniation [12]. Several etiology are considered to underlie the formation of the syrinx, but the main issue was related to disruption of CSF flow between cranial and spinal compartment due to tonsillar and brainstem herniation on the foramen magnum [5], [12], [13]. Intradural pathologies such as arachnoiditis especially on Magendie foramen and cisterna magna also worsen the CSF circulation [5], [13] Reduction in syringomyelia may related to posterior fossa decompression. Geng *et al*. found that early evaluation of the patient shows no difference between two procedures. In time, they found that PFDDP technique is better to reduce syrinx size [13].

The challenge in treating patients with CIM is to make sure that the patients will have the most benefit from the surgery. Several outcome needed to be evaluated to determine the successful of the surgery. Until these days, there is no agreement, on which assessment is the best to evaluate functional outcome CIM patients (Figure 8).

There are no clear indications and surgical technique in managing patient with CIM. General opinion suggests to treat patient with progressive symptoms and syringomyelia with surgery [2]. Individuals without syrinx, with mild symptoms, and no daily lives limitations are better to be observed [14].

Several studies reporting a higher complication rate in in patient treated with PFDDP technique. As we know, disruption of the dura especially on the posterior fossa commonly related to CSF leaks. Williams *et al*. suggest that PFDDP may be performed after failure of

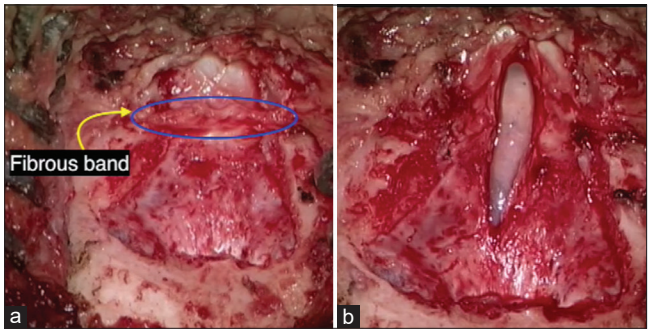

Figure 8: (a) Surgical exposure after performing suboccipital decompression. Fibrous band was mentioned as it will be released and left the dura intact in PFDDS technique. (b) Opening the dura in PFDDP technique that exposes arachnoid and craniocervical junction

PFDDS. This technique may be considered in patient with syringomyelia related to CIM [15], [16].

Regarding the post-operative complications among these two groups, we found that PFDDS group has a lower rate of complications compared to PFDDP. The previous studies also mention that PFDDS technique is safer in the terms of complication. Furthermore, post-operative complication is related to reoperation. Several complications related to surgical procedure in patient with CIM are pseudomeningocele, graft dissolution, irritation, hemorrhage, hydrocephalus, CSF leaks, and infection [17], [18]. Lower length of stay of the patient will influence several outcome of the patient. It may help socioeconomic of the patients and also may reduce infection complication of the patients [17]. One study found that about 35% of patients that undergo PFDDP had post-operative complication compare to only 4% in patient performed PFDDS [3].

Operation time is one of the main issue in surgical technique, as prolonged surgery related to higher chance of infection. High variability in operation time on PFDDS technique may correlate with huge variation of fibrous band adhesion and also surgeon experience to dissect the fibrous band and also control the bleeding. On the experienced hand, PFDDS may be done in shorter surgical duration as it also gives more benefit to the patients [17], [19].

Reoperation in patient with CIM may be due to post-operative complications, or clinical manifestation. CSF leaks that patients may need a further operation to repair the dura and most of it are underwent PFDDP before [20]. As CIM is a chronic progressive disease,

reoperation may also be needed in further time to control the symptoms of the patients. Other surgical approaches such as syringosubdural shunt, arachnoid violation may be used for the next surgery [16].

We also find out about length of stay of CIM patient that underwent surgical management. Effective management with short length of stay must be better choice either for the patient and also the hospital as it benefits for the socioeconomic factor. Besides that, shorter length of stay related to lower chance of hospital-related infection [17].

The purpose of surgery in CIM patient is to expand the posterior fossa compartment and expand the dura to decompress posterior fossa element. Several authors reported about intradural pathological finding that can be evaluated during the surgery and suggest to open the dura to maximally decompress the posterior fossa and reestablished CSF flow [2]. Furthermore, separation of cerebellar tonsils following dura opening may reestablishes CSF free flow from foramen of Magendie [21].

Conclusions

In patients with CIM, we found that the PFDDS technique gives us a lower risk of surgical risk with faster operation time and also lower length of stay which also benefit the patients. There is no superiority between these two surgical procedures regarding reoperation rate, syrinx reduction, and symptoms improvement in CIM patients. Further studies regarding the comparison of these two procedures were needed for this controversy. However, the most important is surgeon's preference and consideration preoperatively for each patient to determine the best surgical approach for the patient.

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