



Comparison of Foramen Magnum Decompression with and without Duraplasty in the Treatment of Adult Chiari Malformation Type I: A Meta-Analysis and Systematic Review

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ABSTRACT

AIM: To include new relevant findings in a systematic review to provide the first comparison of foramen magnum decompression with duraplasty (PFDD) and without duraplasty (PFD) in adult Chiari malformation Type I (CM-I).

MATERIAL and METHODS: We retrospectively searched Web of Science, PubMed, Embase and ClinicalTrials.gov to summarize all relevant published papers on adults. A systemic review was adopted to evaluate clinical or radiological improvement, surgical complications, and reoperation rates between the PFD and PFDD groups.

RESULTS: Nine papers containing information on 497 adult participants met the criteria. PFDD was related to a lower revision rate (RR=2.96, 95% CI: 1.34-6.51, p=0.007) but a higher complication rate (RR=0.35, 95% CI: 0.22-0.55, p<0.00001). No significant difference was noted between PFD and PFDD in terms of overall symptom improvement (RR=0.93, 95% CI: 0.84-1.03, p=0.17) or syringomyelia reduction (RR=0.84, 95% CI: 0.63-1.12, p=0.24). No significant difference in symptom improvement was observed between patients with syringomyelia (RR=0.86, 95% CI: 0.69-1.08, p=0.20) and patients without syringomyelia (RR=0.94, 95% CI: 0.68-1.30, p=0.73).

CONCLUSION: This systematic review of observational studies reveals that PFDD may provide lower revision rates but pose a higher risk than PFD in the management of CM-I in adults. However, PFD is similar to PFDD in clinical and radiological improvements.

KEYWORDS: Chiari malformation Type I; Posterior fossa decompression, Duraplasty, Adult, Meta-analysis

INTRODUCTION

Chiari malformation type I (CM-I) is defined as the cerebellar tonsil moving down 5 mm below the level of the foramen magnum (28). In the general population, the prevalence of CM-I may be up to 1% (40), and 50% - 75% of CM-I patients have syringomyelia (44-46). Posterior

fossa decompression (PFD) is still the first choice to maintain cerebrospinal fluid (CSF) circulation at the level of the foramen magnum (29). Many studies have described the different outcomes of surgical details for adult CM-I: bone decompression alone, removal of the outer layer of the dura (dura-splitting), dural opening with duraplasty (PFDD), or possible tonsil shrinkage (5,6,10,15,18,31-33,36,42).

Extradural decompression usually involves bone removal (5,14,42) or splitting of the dura (15,20,32). In general, PFD does not involve opening the dura mater. Its advantages include fewer complications and a short operation time and hospital stay. PFDD involves duraplasty with or without additional intradural procedures (22), and its theoretical advantages may include low reoperation rates and clinical and radiological improvements. The method can be used to treat intradural lesions (8,29,43). To date, however, no obvious evidence is available to determine which procedure is more suitable for adults with CM-I (3,41,47).

Some meta-analyses have focused on duraplasty in paediatric or mixed populations (3,9,24,25,41,47), while only one obvious heterogeneous meta-analysis is available for adults, where eight of twelve articles were case series without a control group (11). In addition, only four articles included in this heterogeneous meta-analysis compared PFD and PFDD, one of which could not extract the relevant data of the two groups. Distinguishing between children and adults is essential because the characteristics appear to be different in these two populations (11,39). In recent years, many articles comparing PFD and PFDD have been published (5,15,18,32). Therefore, we attempt to include these new findings in a systematic review to guide the latest comparison of duraplasty in adult CM-I.

■ MATERIAL and METHODS

The systematic review was conducted based on the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines (30).

Search Strategy

The Web of Science, PubMed, Embase and Cochrane Library databases were searched by the related MeSH terms or keywords: “decompression” AND “duraplasty” AND “Chiari malformation” AND “adult” from their dates of inception to June 2020. In addition, we applied an English restriction. All relevant abstracts, articles and citations were searched. References of these papers were also manually cross-searched for other potential publications. Since all data extracted are based on previous publications, ethical approval is not required.

Selection Criteria

All articles had to meet the following criteria: 1) CM-I confirmed by MRI, 2) comparison between PFDD and PFD, and 3) an identifiable minimum age of 15 years at surgery. Asymptomatic CM-I patients were also included (5,6). PFD could include bone decompression alone and dura-splitting. PFDD could include duraplasty and additional intradural manipulation procedures. Observational cohort or case-control studies were also incorporated because of the limited number of prospective randomized trials or prospective cohort studies.

The following studies were excluded: 1) animal studies, studies with fewer than ten subjects, editorials, letters, and review articles; 2) multiple papers published related to the same cohort (the most complete study was incorporated to avoid data duplication); and 3) articles without available information for statistical analysis.

Data Extraction and Quality Assessment

All data were extracted from the text, tables and charts of each paper, and any estimates were determined by the data and charts provided. In studies involving subgroups other than CM-I (such as Chiari II) and patient populations, including children and adults, we attempted to extract related data related to patients who met the inclusion criteria for analysis. The following information for each study was independently extracted by two authors (Y.CH. and W.M.): the first author’s name, year of publication, study location, type of the study, surgery, sample size, age or sex, follow-up, overall clinical improvement, clinical improvement with and without syringomyelia, syringomyelia reduction, rates of revision, complications (total complications, CSF-related, infection-related and neurological complications). Any discrepancies were discussed with a third investigator (L.W.).

The primary objectives of this article were to compare (1) overall clinical improvement, (2) syrinx improvement or (3) clinical improvement with and without syringomyelia.

The secondary outcomes of this study were to compare (4) reoperation rates and (5) overall complications.

The quality of articles was evaluated by using a 9-star system from the Newcastle–Ottawa Scale (NOS) (37), and studies with a full score of 9 stars and those with more than 7 stars were included in the systematic review. Because most articles are retrospective and rarely provide details on patient selection for different operative techniques, the comparability score is poor. The difference in scoring the outcomes was particularly significant because results have been inconsistent in different studies. Based on the NOS tool, three studies (31,32,36) scored 8, and the remaining six studies (5,6,10,15,18,42) scored 7. The follow-up time of three studies (5,10,42) was not sufficiently long, the outcome of interest of one study (6) did not appear at the start of the research, and bias was present in the design or analysis of two studies (15,18).

Statistical Analysis

All analyses were performed by Review Manager (version 5.4, www.cochrane-handbook.org), and the outcomes were presented in forest plots. The risk ratio (RR) was used for summary statistics. Both fixed-effects and random-effects models were tested in this study. A random-effects model was used for studies with I^2 values greater than 50%. A funnel plot was used to evaluate publication bias. If substantial heterogeneity was detected, leave-one-out sensitivity analysis was used to qualitatively and quantitatively analyse the possible clinical and methodological reasons.

■ RESULTS

Literature Search

A total of 371 articles were identified by the search strategy. After deleting 116 duplicate studies, the inclusion or exclusion criteria were applied to the titles and abstracts of 239 articles, yielding 22 articles that underwent full-text analysis. The patients in eight articles were all younger than 18 years. Four

articles involved mixed populations or were unsuitable for data extraction. The data of one article overlapped with those in another article, and one was therefore excluded. Finally, nine articles were incorporated (Figure 1). Nine studies (Table I), including eight retrospective observational studies and one randomized study, were published between 2000 and 2019.

Cohort Description

Comparative characteristics and outcomes from each article are presented in Tables I and II, respectively. This study involved a mixed population, but we were able to extract data (31). All other studies included only adults with a minimum age of 15 years. The cohort size of this paper was 497, with 228 (45.9%) patients undergoing PFD and 269 (54.1%) patients undergoing PFDD for the treatment of CM-I. For dural grafts, natural grafts included occipital fascia, cadaveric dura, pericranial autograft, galea aponeurotica, fascia lata and bovine pericardium grafts, while synthetic grafts included DuraGen and pig pericardium grafts (Normal GEN, Guan Hao, Guangzhou, China). Most PFDD approaches involve additional arachnoid dissection and tonsil manipulation (5,6,10,15,32); however, duraplasty without additional intervention was also used (31,36,42). The technique of PFD approaches involves bone decompression

(5,42) and additional dura-splitting (6,10,15,31,32,36); the mean ages of patients at the time of surgery were 36.4 and 37 years, respectively. The proportion of male patients treated by PFD and PFDD ranged from 19.5% to 75% and 12.5% to 100%, respectively. The average follow-up ranged from 6 months to 8 years.

Primary Outcomes

Overall Clinical Improvement

Nine studies were identified for an analysis of overall symptom improvement in patients. The incidence rates of overall symptom improvement after PFD and PFDD were 180 of 228 (78.9%) and 220 of 269 (81.8%), respectively. No significant difference was found between the two subgroups, with a pooled RR of 0.93 (95% CI: 0.84-1.03, $p=0.17$) (Figure 2A). The degree of heterogeneity was moderate ($I^2 = 33\%$, $p=0.17$), and a fixed-effects model was used. The detailed results of subgroup analyses stratified by types of PFD (bone decompression alone vs. duraplasty and dura-splitting vs. duraplasty) were analysed. Four studies were incorporated for a pooled analysis of symptom improvement in patients with or without syringomyelia. In the subgroup with syringomyelia, no

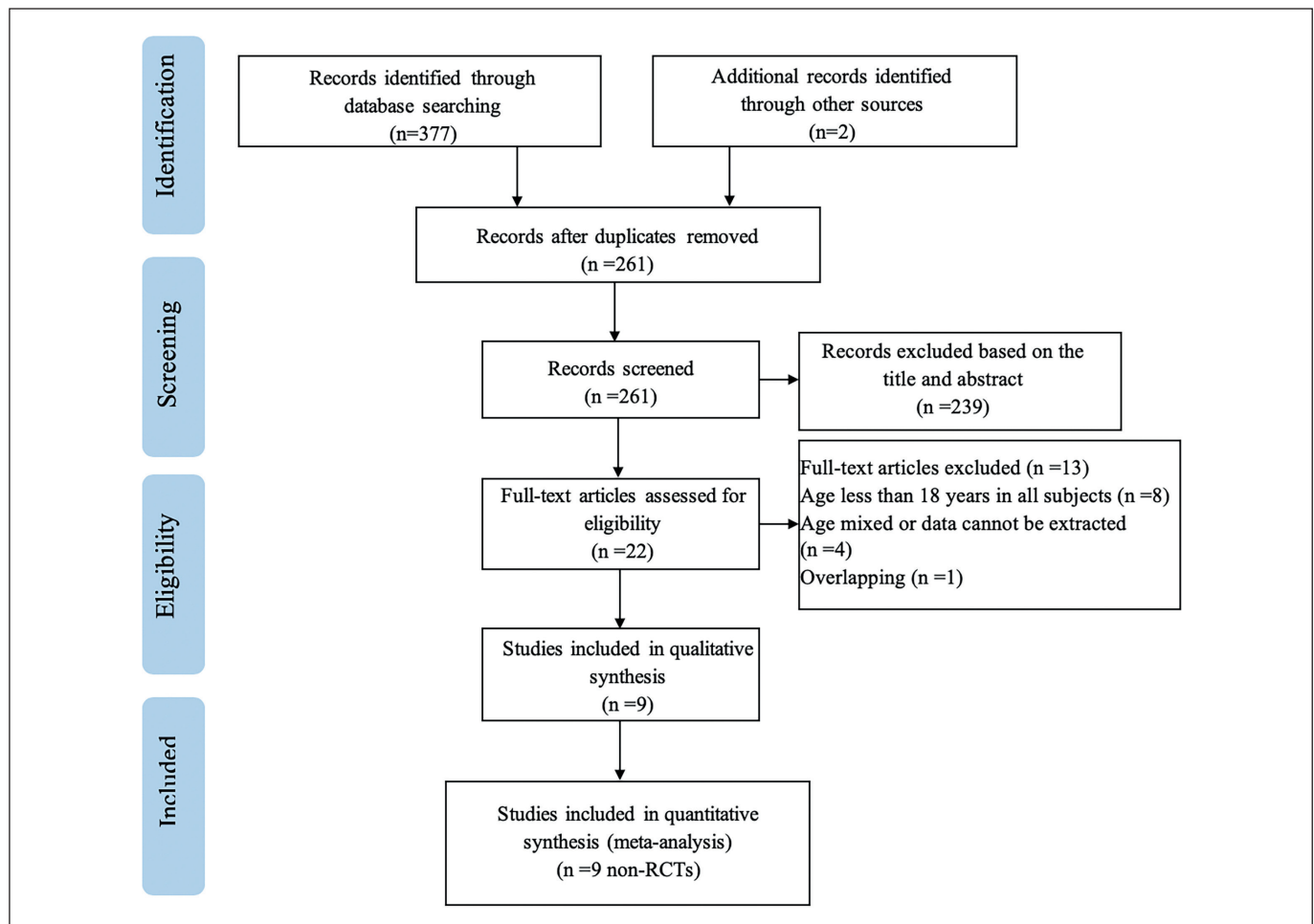


Figure 1: Flow chart of study identification.

Table I: Study Characteristics and Basic Demographics

Authors & Year	Country	NOS	Study Design	No. of Pts	No. of Pts Treated		Dural Graft		Mean Age (yrs)		Sex (M/F)		Mean Follow-Up (mos)	
					PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD
Munshi et al., 2000* (31)	USA	8	R, OS	27	11	16	Natural		38	39.3	4/7	2/14	9 to 8 yearst	
Erdogan et al., 2010(10)	Turkey	7	R, RS	27	12	15	Natural		31.58	25.86	9/3	15/0	NA†	
Romero and Pereira, 2010 (36)	Brazil	8	R, OS	16	6	10	Natural		41	40	3/3	3/7	9-24†	
Yilmaz et al., 2011 (42)	Turkey	7	R, OS	82	24	58	Natural		31	38.9	36/46†		6-12†	
Chotai and Medhkour, 2014 (6)	USA	7	R, OS	41	29	12	Natural, synthetic		33.8†		8/33†		14.7 †	
Gurbuz et al., 2015 (18)	Turkey	7	R, OS	39	18	21	NA		36†		13 /26†		43†	
Chen et al., 2017 (5)	China	7	R, OS	103	33	70	Natural, synthetic	40.8	40.6	9/24	23/47		1 year†	
Geng et al., 2018 (15)	China	7	R, OS	49	17	32	Synthetic	39.00	42.13	6/11	7/25	39.24	43.03	
Oral et al., 2019 (32)	Turkey	8	R, OS	113	78	35	Natural	19-66	22-56	22/56	21/14		1 year†	
Total				497	228 (45.9%)	269 (54.1%)								

NA: Not applicable, OS: Observational study, RS: Randomized study, Pts: Patients, R: Retrospective, M: Male, F: Female.
 * The reported value of <15-year-old was excluded. † Authors reported the value for the entire cohort only.

Table II: Postoperative Outcomes of Studies with Rates Expressed as Patient Number/Available Cohort

Authors & Year	Overall Clinical Improvement		Syrinx Improvement		Postop Complications		CSF-Related		Infection-Related		Neurological complications		Hospital stays (d)		Revision	
	PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD
Munshi et al., 2000	8/11	13/16	3/6	6/6	1/11*	10/23*	0	NA	0	NA	0	NA	NA	NA	2/11	0/16
Erdogan et al., 2010	10/12	11/15	4/4	10/11	0/12	3/15	0/12	3/15	0/12	0/15	0/12	0/15	NA	NA	1/12	0/15
Romero et al., 2010	4/6	9/10	2/4	5/5	1/6	5/10	0/6	3/10	1/6	1/10	0/6	1/10	NA	NA	NA	NA
Yilmaz et al., 2011	19/24	52/58	16/19	41/45	2/24	7/58	0/24	3/58	1/24	2/58	1/24	2/58	NA	NA	2/24	0/58
Chotai et al., 2014	26/29	9/12	0	5/5	1/29	4/12	0/29	1/12	1/29	1/12	1/29	2/12	3.1	4.1	0/29	1/12
Gurbuz et al., 2015	11/18	17/21	1/8	12/13	1/18	6/21	0/18	2/21	1/18	4/21	0/18	0/21	NA	NA	4/18	3/21
Chen et al., 2017	23/33	57/70	NA	NA	8/33	23/70	1/33	0/70	3/33	21/70	2/33	0/70	NA	NA	2/33	0/70
Geng et al., 2018	8/17	24/32	NA	NA	0/17	10/32	0/17	2/32	0/17	7/32	0/17	1/32	10	12.6	NA	NA
Oral et al., 2019	71/78	28/35	33/40	14/17	4/78	10/35	0/78	5/35	4/78	5/35	0/78	0/35	NA	NA	7/78	0/35
Overall (%)	180/228(20/269)	59/81 (81.8%)	88/97 (72.8%)	90.7%	17/217 (7.8%)	68/253 (26.9%)	1/217 (0.4%)	19/253 (7.5%)	11/205 (5.3%)	41/238 (17.2%)	4/217 (1.8%)	6/253 (2.4%)	NA	NA	18/205 (8.8%)	4/227 (1.8%)

* Reported value for overall cohort, we cannot exclude the reported value of < 15-year-old.

significant difference was noted (RR 0.86, 95% CI: 0.69-1.08, p=0.20) (Figure 2B). No significant difference was identified in the subgroup without syringomyelia (RR 0.94, 95% CI, 0.68-1.30, p=0.73) (Figure 2C), and the outcomes were found to have no heterogeneity ($I^2=0\%$, p=0.73). No significant difference in clinical improvement was identified between the bone decompression alone group and PFDD group (RR 0.87, 95% CI: 0.73-1.03, p=0.1) or the dura-splitting group and PFDD group (RR 1.0, 95% CI: 0.87-1.15, p=0.99) (Figure 3).

Syringomyelia Reduction

The incidence rates of syringomyelia reduction after PFD and PFDD were 59 of 81 (72.8%) and 88 of 97 (90.7%), respectively. No significant difference was noted between the two subgroups, with a pooled RR of 0.84 (95% CI: 0.63-1.12, p=0.24) based on six studies (Figure 4A). The degree of heterogeneity was high ($I^2=59\%$, p=0.24), and a random-effects model was applied. In the sensitivity analysis, when we deleted one study (18), lower heterogeneity was found (RR 0.93, 95% CI, 0.79-1.09, $I^2=7\%$, p=0.37). This change might be caused by the limited sample size in the article, which affected the heterogeneity of the outcome of syrinx improvement.

Secondary outcomes

Revision Operation

The incidence rates of revision operations after PFD and PFDD were 18 of 205 (8.7%) and 4 of 227 (1.7%), respectively. PFD was related to a significantly different possibility of revision surgery compared with PFDD, with an RR of 2.96 (95% CI: 1.34-6.51, p=0.007) based on 7 articles (Figure 4B).

Complications

The incidence rates of overall complications after PFD and PFDD were 17 of 217 (7.8%) and 68 of 253 (26.9%), respectively. PFDD was related to a significantly higher incidence of overall complications than PFD (RR 0.35, 95% CI: 0.22-0.55, p<0.00001) based on 8 articles (Figure 4C). Furthermore, PFDD was related to significantly higher incidence rates of CSF-related (RR 0.26, 95% CI: 0.11-0.61, p=0.002) complications compared with PFD based on 8 studies (Figure 5A) and infection-related (RR 0.35, 95% CI 0.18-0.68, p=0.002) complications compared with PFD based on 7 articles (Figure 5B). In addition, PFDD was not associated with a significantly different possibility of neurological

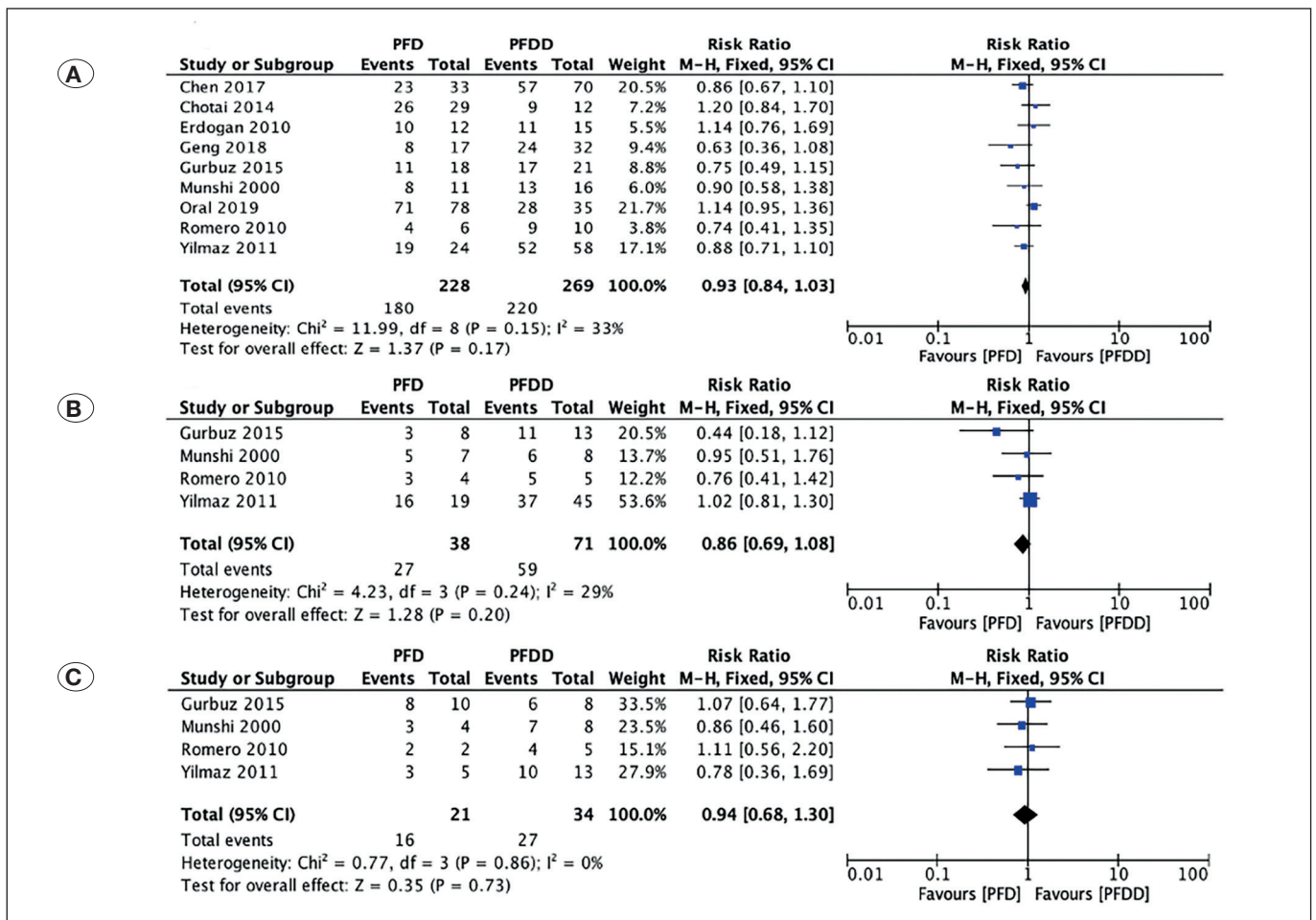


Figure 2: Forest plots comparing improvements in overall clinical status with PFD versus PFDD in the treatment of adult CM-I; no significant difference in clinical improvement was found among all subgroups (A), in the syringomyelia subgroup (B), and the without syringomyelia subgroup (C).

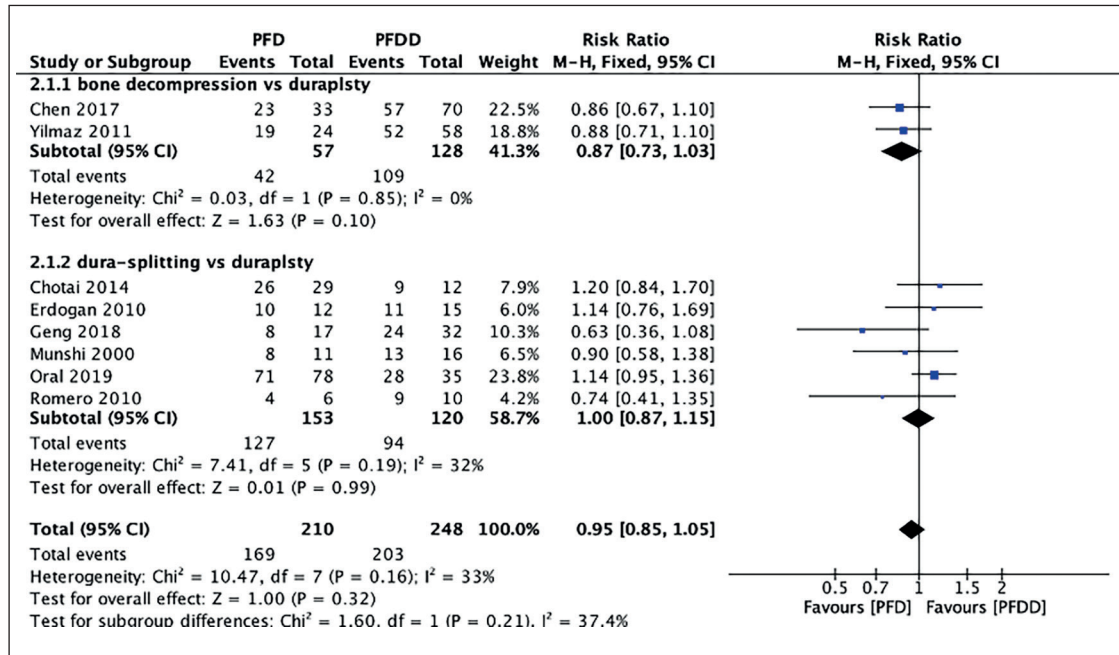


Figure 3: Forest plots comparing improvements in overall clinical status with PFD versus PFDD in the treatment of adult CM-I in subgroup analyses between bone decompression vs. duraplasty and duralsplitting vs. duraplasty. No significant difference in clinical improvement was found between the bone decompression alone group and PFDD group or between the dura-splitting group and PFDD group.

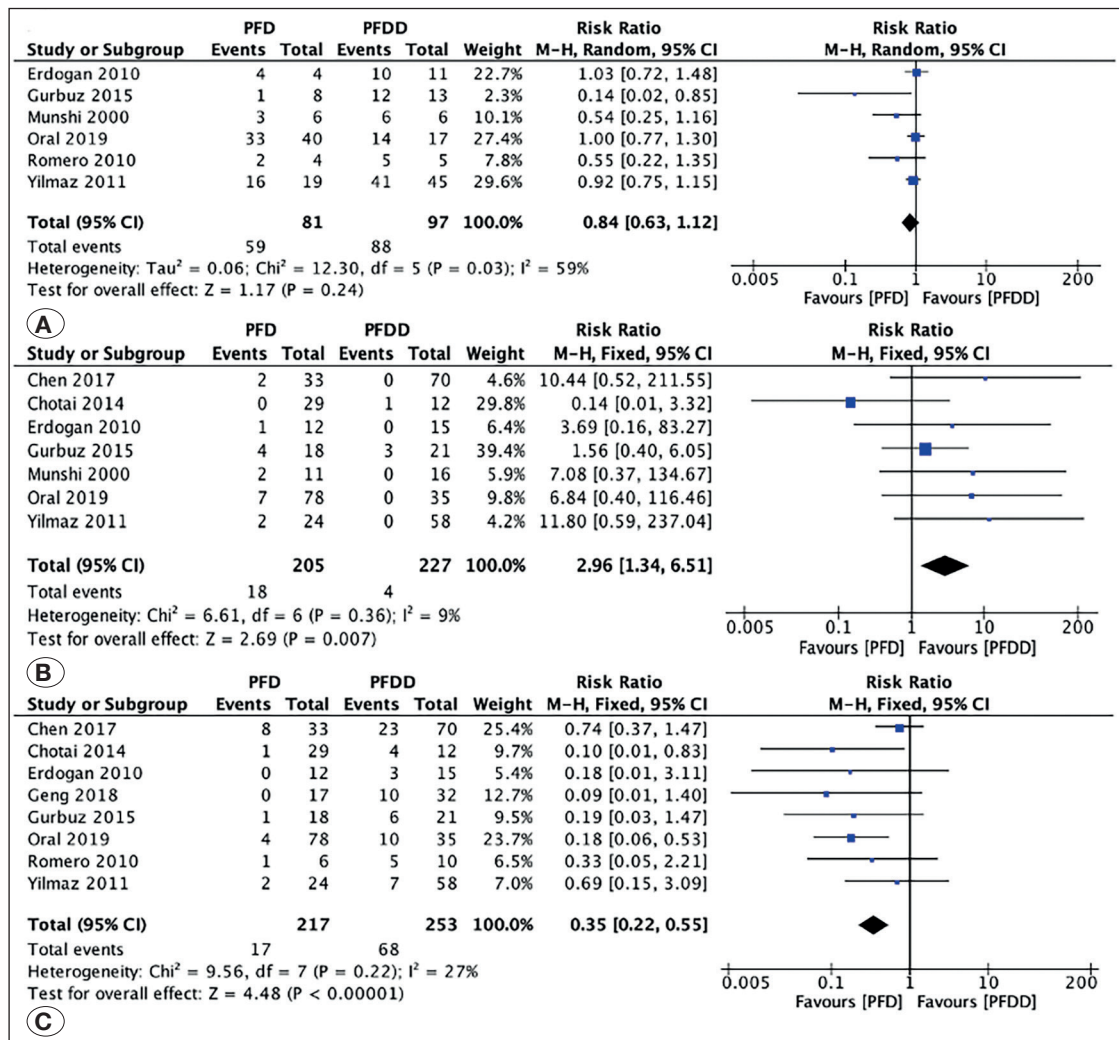


Figure 4: Forest plots comparing surgical outcomes of PFD versus PFDD in the treatment of adult CM-I with respect to syringomyelia improvement (A), revision surgery (B), and overall complications (C). Significant differences in revision surgery and overall complications were found between the PFD group and PFDD group except for syrinx improvement.

complications (RR 1.01, 95% CI: 0.38-2.69, p=0.98) compared with PFD based on 8 articles (Figure 5C).

Publication Bias

Funnel plots were conducted for each outcome. No significant asymmetry was noted; thus, no evidence of publication bias was observed.

Sensitivity Analysis

Sensitivity analysis was conducted during the review to assess the impact of individual articles on the results of the pooled analysis. If one article was omitted at a time, the results were considered highly deterministic if the overall results did not change. After each study was omitted, no significant change (except syringomyelia improvement) in result trend directions was observed.

Sensitivity analysis revealed that study (18) was the origin of the statistical heterogeneity in the systematic review for syringomyelia reduction. When this study was removed, we found low heterogeneity in the five remaining studies (RR 0.93,

95% CI: 0.79-1.09, I²=7%, p=0.37), and a meta-analysis of these five trials still demonstrated no significant difference in syringomyelia improvement between PFD and PFDD. This changed heterogeneity might be related to the limited sample size of this study. In this study, significant differences in syringomyelia improvement were found between the two subgroups, while the bias of the design and analysis was obvious since only 21 patients with syringomyelia were included.

DISCUSSION

Some meta-analyses and systematic reviews have suggested that PFDD is superior to PFD in terms of overall clinical improvement (13,25,41,47), while some studies have shown that PFDD is better at least for patients with syringomyelia (24). However, some systematic reviews have reported no significant difference between the two approaches (3,9,11,33). In this systematic review, the overall clinical improvement with PFD was not inferior to that with PFDD. In the subgroup analysis of different types of PFD, we found no significant

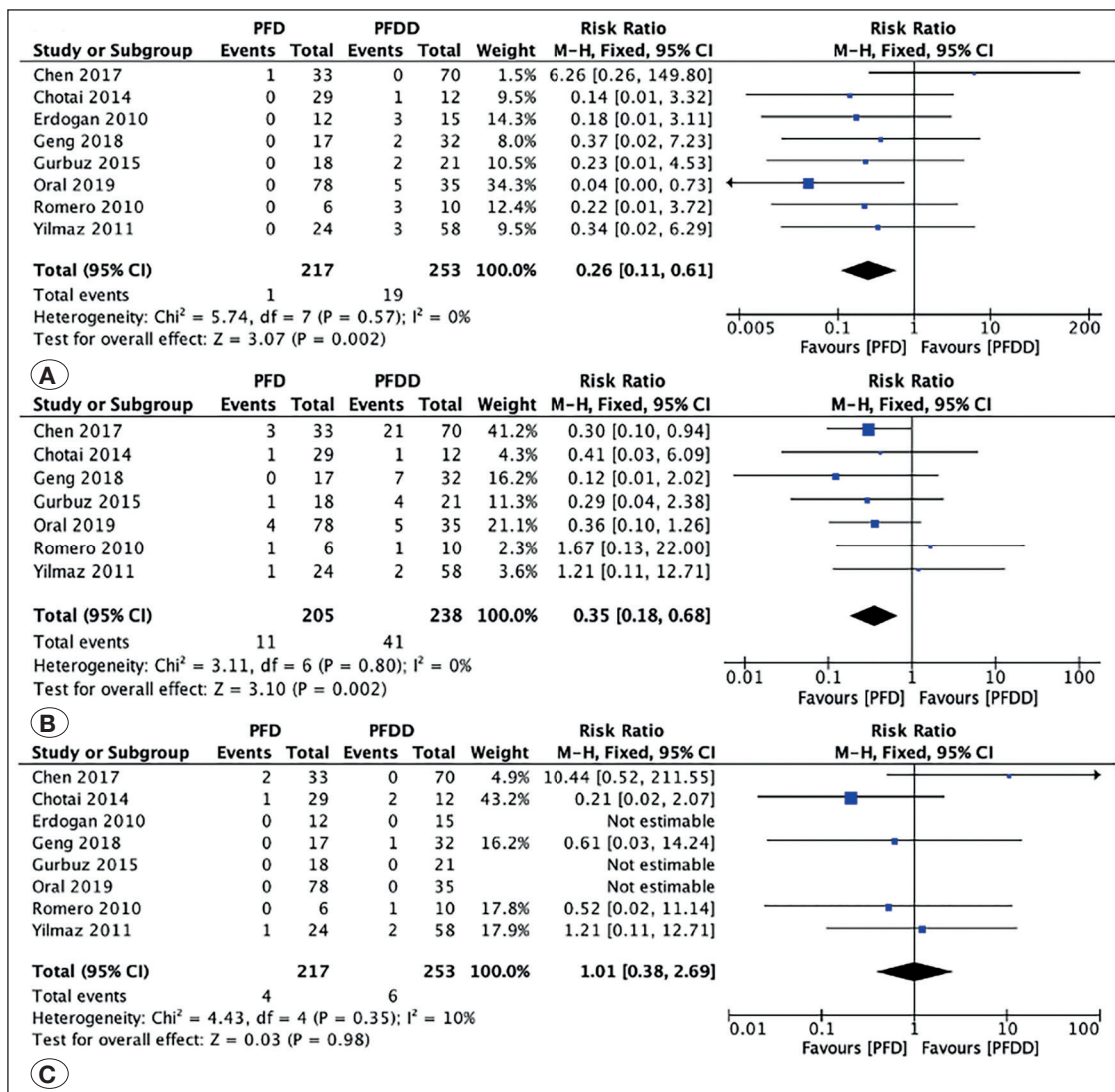


Figure 5: Forest plots comparing PFD versus PFDD in the treatment of adult CM-I in terms of CSF-related (A), infection-related (B), and neurological complications (C). Significant differences in CSF-related complications and infection-related complications were found between the PFD group and PFDD group but not for neurological complications.

differences between the two groups (bone decompression vs. duraplasty and dura-splitting vs. duraplasty). This outcome is similar to previously reported results (38). Furthermore, when stratified by syringomyelia status, no significant difference was observed in terms of clinical improvement, indicating that syringomyelia might not be a risk factor for clinical improvement. Our results must be treated with caution because the design bias, limited sample size, lack of an agreed-upon assessment scale and uncontrollable clinical effect selection bias complicate interpretation of the study's validity. Overall symptom improvement refers to symptomatic improvement from the preoperative state to the postoperative state. The symptoms of adult CM-I patients are recognized to vary substantially (26), and due to this heterogeneity of CM-I symptoms, comparisons before and after surgery are difficult.

Some studies also suggest that the decrease in syringomyelia is not as high as the overall symptom improvement (18,42). Recently, Perrini et al. (34) concluded that patients undergoing dura-splitting had a lower rate of syringomyelia reduction than those undergoing PFDD. However, the literature included in this comparison obviously affected the systematic review due to confounding factors. Some people believe that CM-I is associated with embryonic mesodermal somite occipital lobe dysplasia and encroachment on the posterior fossa (23), but others believe that intradural lesions such as cerebellar tonsillar herniation and other possible arachnoid adhesions are associated with CM-I (7), which may be the cause of syringomyelia. However, given the narrow subarachnoid space at the craniocervical junction, PFD can improve CSF circulation without opening the dura mater, which is sufficient to reduce the impact of intradural lesions (4,38). The lack of statistical significance in the improvement rate index of this meta-analysis may be due to the limited sample size, the time point of the imaging follow-up or the definition of imaging improvement reported in the current literature (1). Recently, in a prospective study on adolescents (mean age 13.8 years) published by Jiang et al., the authors also suggested that PFD is the best choice for CM-I (21).

PFDD includes duraplasty and intradural manipulation according to the surgeon's preference (16,17,26). Generally, this is a more aggressive approach and is expected to obtain a better surgical outcome, which explains the longer length of stay after PFDD compared with PFD. Two observational studies (6,15) suggested that in adult CM-I treatment, the PFDD method required more time than the PFD method in terms of hospital stay. Likewise, the higher incidence of CSF- and infection-related complications after duraplasty than after PFD may also be due to the more extensive nature of PFDD. CSF-related complications include hydrocephalus, CSF leakage, pseudomeningocele, arachnoid scar formation and meningitis, resulting in CSF circulation disorders and foreign-body reactions (9,41). In general, most complications are associated with CSF exposure to blood, muscle cells and dural transplantation. Therefore, PFD can reduce the incidence of complications.

According to comparative studies, PFD is related to a higher rate of revision surgery than PFDD. Unfortunately, the timing of each revision is uncertain in this meta-analysis, which

complicates comprehensive interpretation of these outcomes because the indications for revision surgery, including short-term complications or long-term follow-up symptoms, are not improved (43). If the difference in the revision rate with the PFDD or PFD approach is related to short-term or long-term follow-ups, the different follow-up time points of each article will engender a selection bias for these outcome indicators. Similarly, for outcome indicators such as overall clinical improvement, different follow-up time points can also cause the above situation. Regardless of the technique, some patients still had symptoms after decompression of the posterior fossa. According to our experience, the causes may be misdiagnosis, persistent symptoms, or surgery introducing a previously nonexistent mechanism (43). If only PFD is performed, duraplasty and even additional intradural manipulation can be considered in revision surgery, but this approach may affect the number of reoperations because of clinical failure in the PFD group. However, even if sufficient PFDD is performed for the first time, patients' symptoms are not improved, and further surgical decompression is usually not feasible.

As the method to evaluate the need for duraplasty, intraoperative ultrasound has been tried by many authors (27), and in a series of recent prospective studies, intraoperative MRI was also used (2). The results showed that CSF circulation was improved in the prone position during the operation; thus, CSF circulation did not improve significantly after PFD. Unfortunately, this observation further illustrates the intraoperative bias. Therefore, the surgical treatment of Chiari I should continue to be adjusted according to preoperative clinical (12,35,42) and radiological criteria (19,42,43) rather than intraoperative radiological criteria. In addition, the problem of distinguishing different subgroups of CM-I patients remains, and the best indication for PFD or PFDD remains to be determined.

■ CONCLUSION

Both PFD and PFDD can achieve satisfactory results in the treatment of adult CM-I. This systematic review showed that PFDD was related to lower revision rates and more postoperative complications than PFD. However, no obvious significant difference in clinical results or the syrinx improvement rate was found between the two techniques. We suggest that when explaining the advantages and disadvantages of the two surgical methods to patients in detail, PFD should be recommended first.

■ AUTHORSHIP CONTRIBUTION

Study conception and design: CY

Data collection: MW

Analysis and interpretation of results: CY

Draft manuscript preparation: WM

Critical revision of the article: WL, XW

Other (study supervision, fundings, materials, etc.): FJ

All authors (CY, MW, WL, XW, FJ) reviewed the results and approved the final version of the manuscript.

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