



Posterior fossa decompression with duraplasty in Chiari malformation type 1: a systematic review and meta-analysis

Sharon Ka Po. Tam¹ · Andrew Brodbelt² · Paolo A. Bolognese³ · Mansoor Foroughi⁴

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Abstract

Background Surgery for symptomatic Chiari type I malformation (CM-I) patients include posterior fossa decompression (PFD) involving craniectomy with or without dural opening, and posterior fossa decompression with duraplasty (PFDD). This review aims to examine the evidence to aid surgical decision-making.

Methods A medical database search was expanded to include article references to identify all relevant published case series. Animal studies, editorials, letters, and review articles were excluded. A systemic review and meta-analysis were performed to assess clinical and radiological improvement, complications, and reoperation rates.

Results Seventeen articles, containing data on 3618 paediatric and adult participants, met the inclusion criteria. In the group, 5 papers included patients that had the dura left open. PFDD is associated with better clinical outcomes (RR 1.24, 95% CI, 1.07 to 1.44; $P = 0.004$), but has a higher complication rate (RR 4.51, 95% CI, 2.01 to 10.11; $P = 0.0003$). In adults, clinical outcomes differences did not reach statistical significance ($P = 0.07$) but re-operation rates were higher with PFD (RR 0.17, 95% CI 0.03 to 0.86; $P = 0.03$), whilst in children re-operation rates were no different (RR 0.97, 95% CI 0.41 to 2.30; $P = 0.94$). Patients with a syrinx did better with PFDD ($P = 0.02$). No significant differences were observed concerning radiological improvement.

Conclusions In the absence of hydrocephalus and craniocervical region instability, PFDD provides better clinical outcomes but with higher risk. The use of PFD may be justified in some cases in children, and in the absence of a syrinx. To help with future outcome assessments in patients with a CM-I, standardization of clinical and radiological grading systems are required.

Trial registration: not required

Keywords Chiari · Treatment · Duraplasty · Syringomyelia · Dural patch

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✉ Sharon Ka Po. Tam
sharontam31@outlook.com

¹ Leighton Hospital, Mid Cheshire Hospitals NHS Foundation Trust, Crewe, UK

² The Walton Centre, NHS Foundation Trust, Liverpool, UK

³ Mount Sinai South Nassau, Chiari Neurosurgical Institute, Oceanside, NY, USA

⁴ Royal Sussex County Hospital, Brighton and Sussex University Hospitals NHS Trust, Brighton, UK

Introduction

A Chiari malformation (CM) is the downward displacement of the cerebellar tonsils through the foramen magnum into the upper cervical spinal canal [1]. With the increasing availability of neuroimaging, up to 1% of the general population is being diagnosed with Chiari malformation type 1 (CM-I) [22]. Most individuals with CM-I remained asymptomatic, but up to 7% of those diagnosed radiologically will develop a spectrum of symptoms and signs, ranging from headaches and nausea to brainstem compression [16].

Surgery can be effective in the treatment of patients with symptomatic CM-I with or without syringomyelia. There is much debate over the ideal surgical approach, and one area of controversy relates to patching the dura. A posterior fossa decompression involving craniectomy with or without opening the dura (PFD) appears to be lower-risk, have shorter operating times, is technically easier, and is becoming more

Table 1 Characteristics of studies

Authors and year	Country	Interventions			Age (years)		Follow-up	
		<i>N</i>	PFD	PFDD	PFD	PFDD	PFD	PFDD
Munshi et al., 2000 [19]	USA	34	11	23	38	29.9	9 months to 8 years	
Ventureyra et al., 2003 [27]	Canada	16	8	8	10.5		6 months	
Navarro et al., 2004 [21]	USA	96	69	27	8.2		0.17 to 9.8 years (2.3 years)	
Yeh et al., 2006 [30]	USA	130	40	90	5.9		20 months	
Galarza et al., 2007 [8]	USA	41	20	21	10		1 to 10 years (21 months)	
Mutchnick et al., 2010 [20]	USA	120	56	64	11.1		6 months	
Romero et al., 2010 [25]	Brazil	16	6	10	40.6		9 months to 2 years	
Litvack et al., 2013 [18]	USA	110	63	47	8.3	10.4	418 days	561 days
Chotai et al. 2014 [6]	USA	41	29	12	33.8		6 to 12 months	
Lee et al., 2014 [17]	USA	65	29	36	8.9	9.9	12 months to 36 months	
Shweikeh et al. 2014 [26]	USA	2649	1593	1056	9.8	10.9	NA	
Gurbuz et al., 2015 [12]	Turkey	25	12	13	36		44 months	
Awad et al., 2016 [3]	Egypt	20	10	10	32.3	34.2	1 year	
Chen et al., 2017 [5]	China	103	33	70	40.8	40.6	1 year	
Raza-Knight et al. 2017 [24]	UK	54	16	38	8.7		NA	
Grahovac et al. 2018 [9]	USA	16	10	6	1.7		3 to 10 years (5.6 years)	
Jiang et al. 2018 [13]	China	82	40	42	13.6	14.0	35.2 months	36 months

popular in paediatric patients. In the PFD group, the dura is not opened at all, opened and closed primarily, or opened and left open. A number of, often small, studies have compared PFD with posterior fossa decompression with duraplasty (PFDD). To date, no consensus has been reached regarding the optimal surgical management for patients with CM-I.

The aim of this study is to critically evaluate the published literature comparing PFD and PFDD, examining clinical and radiographic improvements, complications, and reoperation rates. Information gathered from this study will be used to help guide surgical decision-making in the treatment of patients with CM-I.

Methods

Literature search strategy

This systematic review and meta-analysis was conducted based on the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines. Electronic searches were performed using Embase, PubMed, Cochrane and Web of Science using the following combined terms: ‘Chiari malformation’, ‘duraplasty’, and ‘bony decompression’. We applied an English language restriction. To further identify all potentially relevant studies, we manually searched reference lists from all the retrieved articles. No

ethical approval was required, as all data extracted were based on previous publications.

Study selection

The primary outcomes of this study were to compare (1) clinical improvement, (2) radiological improvement, (3) overall complications, and (4) reoperation rates in CM-I patients who underwent either PFD or PFDD. Inclusion criteria were all patient series with symptomatic CM-I associated with posterior fossa volume mismatch in the absence of hydrocephalus and instability of the craniocervical region. No age restriction was applied. Retrospective and prospective cohort studies were included because of the paucity of prospective randomized trials. Animal studies, editorials, letters, and review articles were excluded from this study. Articles with accumulation of study groups were reviewed, and the most completed studies were selected to avoid data duplication.

Data collection

Two independent investigators (SKPT and MF) evaluated all studies. The following data were extracted from each report: (1) study characteristics, (2) patient characteristics, (3) types of treatment, and (4) outcomes. Any discrepancies were evaluated by repeat review and resolved by consensus. Attempts were made to contact the study’s authors if further clarification

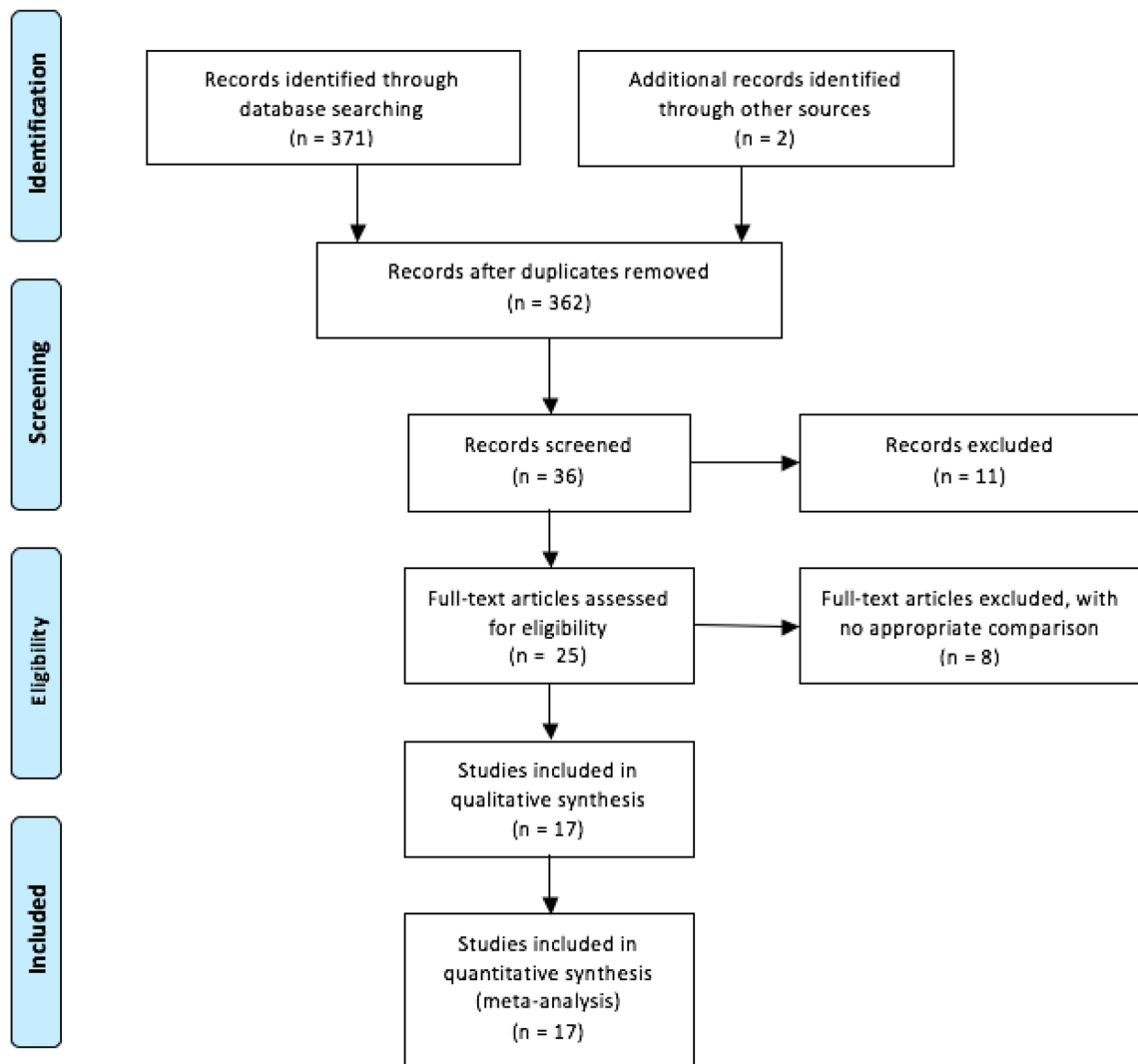


Fig. 1 Flow chart of identification of studies

was required. Clinical improvement was not always consistently defined, but included patient-reported symptom improvement, reduced pain, return to work, reduced analgesic

medication, and neurological improvement. Radiographic improvement was only described in patients with an associated syrinx, and was assessed using magnetic resonance imaging

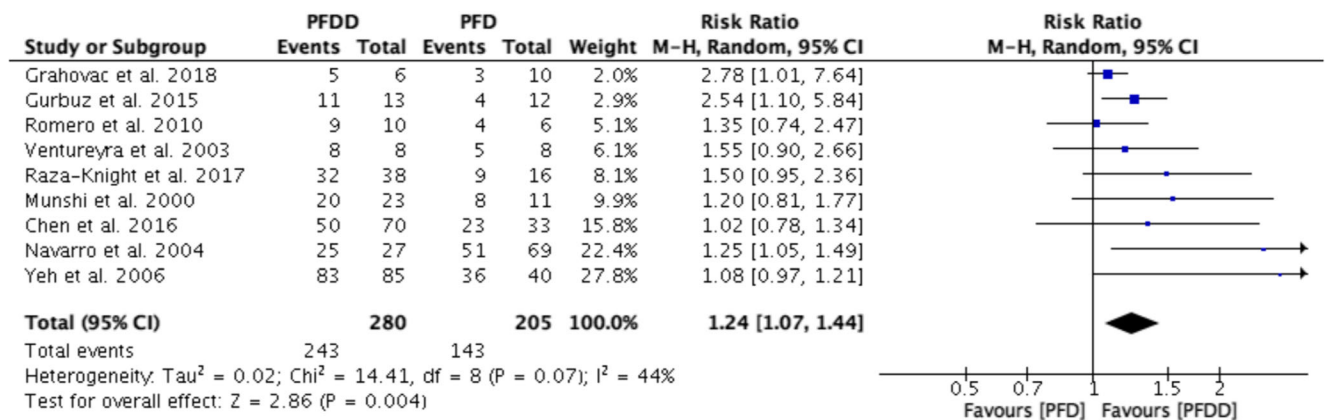


Fig. 2 Clinical improvement

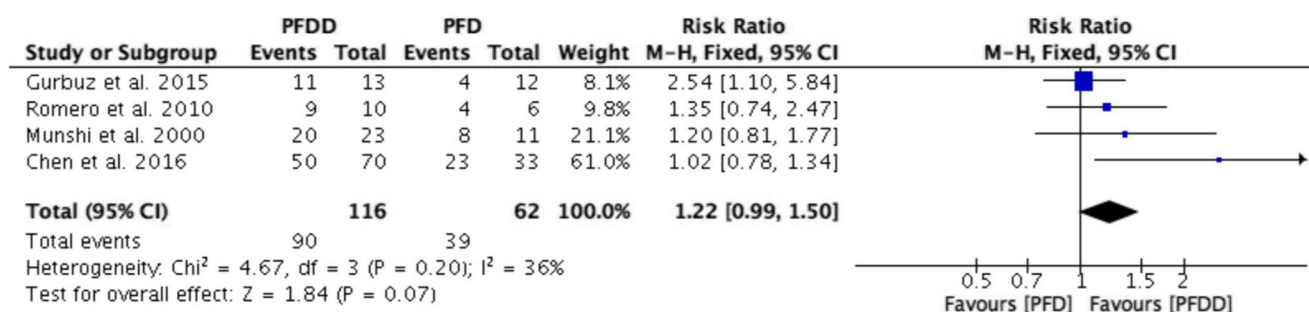


Fig. 3 Clinical improvement—adults

(MRI) to determine the degree of syrinx regression (defined as a decrease in maximal syrinx diameter). The quality of evidence was evaluated using the Newcastle-Ottawa tool.

Statistical analysis

All statistical analyses were conducted using Review Manager (version 5.3.5, the Cochrane Collaboration, 2014), and the results were displayed in forest plots. We calculated an overall relative risk (RR) to assess the ratios of probabilities of clinical improvement, radiological improvement, complications, and reoperation rates in CM-I patients treated with PFD and PFDD. Ninety-five percent confidence intervals were calculated for all categorical outcomes. Pool participant data were tested in a fixed-effects or random-effects model, selected depending on the degree of heterogeneity between studies. Cochran's chi-squared test (χ^2) and I^2 test were used to analyse the between-study heterogeneity. A random-effects model was used for studies with $P < 0.10$ and $I^2 > 50\%$. Publication bias was assessed using funnel plots. Sensitivity analysis was conducted to test the robustness of the measured outcomes.

Results

Characteristics of the studies

A summary of the characteristics of the included studies is presented in Table 1. A total of 371 studies were

identified from electronic searches, of which 17 articles met the inclusion criteria and were included (Fig. 1). The 17 studies, including 16 retrospective cohort studies and one prospective cohort study, were published between 2000 and 2018. A total of 2045 patients underwent PFD and 1573 patients underwent PFDD. The quality of individual studies was assessed using the Newcastle-Ottawa tool, with 13 studies scoring 8 and four studies scoring 7. Two of these studies did not have a long enough follow-up for outcomes to occur (Shweikeh et al. 2014 and Raza-Knight et al. 2017), and two of these studies' outcome of interest were not present at start of study (Navarro et al., 2004 and Chotai et al. 2014). Studies that scored less than 7 were excluded from this review.

Quantitative synthesis

Clinical improvement

A total of 9 studies, including 280 patients who underwent PFDD and 205 patients who underwent PFD, were identified for the analysis of overall clinical improvement (Fig. 2). A significant difference was observed between the two groups, with a pooled RR 1.24 (95% CI, 1.07–1.44; $P = 0.004$). The degree of heterogeneity was moderate ($I^2 = 44\%$; $P = 0.07$), and a random-effects model was used (Fig. 2). In the sensitivity analysis, PFDD was associated with better overall clinical improvement, with no significant between-study heterogeneity.

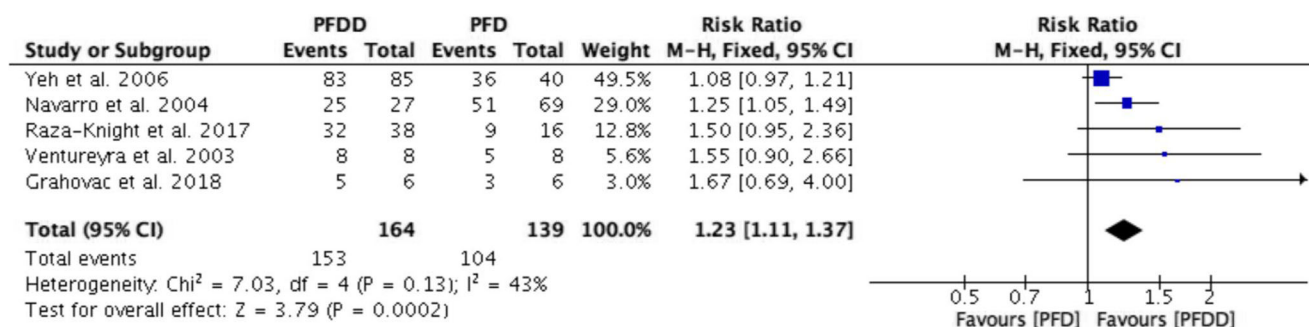


Fig. 4 Clinical improvement—children

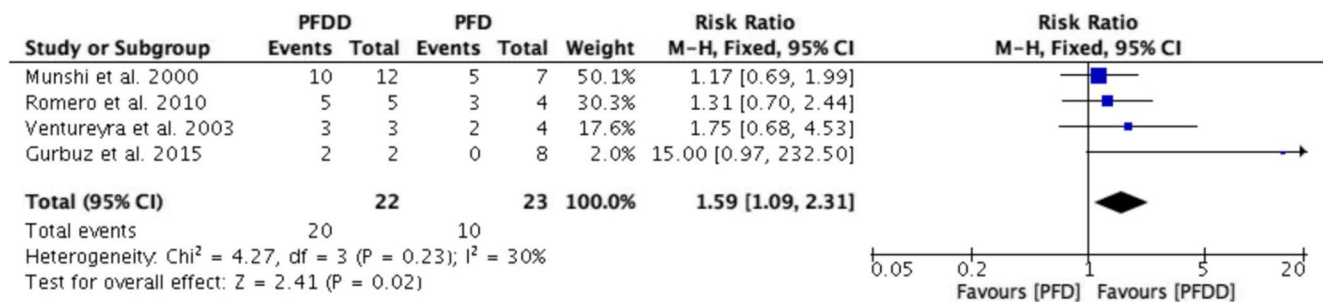


Fig. 5 Clinical improvement—with syringomyelia

The subgroup analysis of adults did not reach significance (RR 1.22, 95% CI, 0.99–1.50; $P = 0.07$) (Figure 3). The analysis of children did reveal a significant difference in clinical improvement favouring PFDD (RR 1.23, 95% CI, 1.11–1.37; $P = 0.0002$) (Fig. 4). Four studies were identified for a pooled analysis of clinical improvement in patients with and without syringomyelia. In patients with a syrinx, a significant difference was observed favouring PFDD (RR 1.59, 95% CI, 1.09–2.31; $P = 0.02$) (Fig. 5). In patients without a syrinx, no significant difference was observed (RR 1.07, 95% CI, 0.82–1.39; $P = 0.61$), with low heterogeneity ($I^2 = 0\%$, $P = 0.90$) (Fig. 6).

Radiological/imaging improvement

Seven studies, including 105 patients who underwent PFDD and 84 patients who underwent PFD, were identified for the analysis of radiographic improvement. There was no significant difference in radiological improvement between the two groups (RR 1.15, 95% CI, 0.85–1.56; $P = 0.36$), including the subgroup analysis into adult and children (Fig. 7). A random-effects model was used. In the sensitivity analysis, no significant difference in radiological improvement was observed between the two groups (Figs. 8 and 9).

Complications

Nine studies, including 1361 patients who underwent PFDD and 1850 patients who underwent PFD, reported

the overall complication rates. Complications included CSF leak, aseptic meningitis, pseudomeningocele, seroma collections, and wound infection. A significant difference between the two groups (RR 4.51, 95% CI, 2.01–10.11; $P = 0.0003$) was observed which persisted in subgroup analysis (Figs. 10, 11, 12, and 13). A random-effects model was used due to the high levels of heterogeneity ($I^2 = 83\%$, $P < 0.00001$). In the sensitivity analysis comparison, a stable outcome was observed.

Reoperations

A total of 10 studies, including 1411 patients who underwent PFDD and 1914 patients who underwent PFD, reported the rate of reoperations (Figs. 14, 15, and 16). No significant difference was observed between the two groups (RR 0.69, 95% CI, 0.29–1.65; $P = 0.4$). In the sensitivity analysis comparison, a stable outcome was observed. Only in adults was there a significant difference with higher reoperation rates after PFD (RR 0.17, 95% CI, 0.03–0.86; $P = 0.03$), with low heterogeneity ($I^2 = 0\%$, $P = 0.66$) (Fig. 15).

Sensitivity analyses

Sensitivity analyses were performed during the review process to evaluate the effect of individual studies on the outcomes of pooled analyses. One study was omitted at a time and if the overall results showed no changes, the

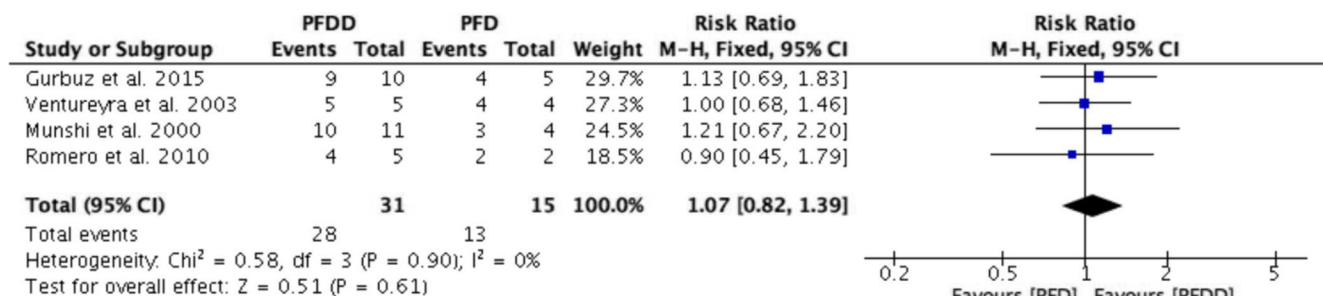


Fig. 6 Clinical improvement—without syringomyelia

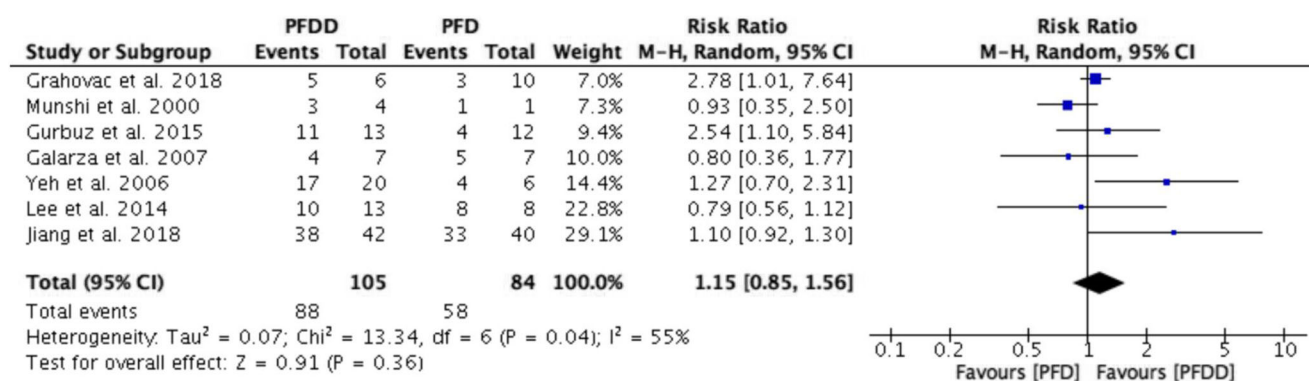


Fig. 7 Radiological improvement

results were considered as having a high degree of certainty.

Publication bias

Each outcome measure was assessed using funnel plot to identify any potential publication bias. The funnel plots did not show evidence of significant asymmetry.

Discussion

This systematic review and meta-analysis were conducted to compare two surgical techniques in CM I patients, namely, PFD and PFDD. In this study, CM-I patients who underwent PFDD achieved a better overall clinical improvement compared with PFD, especially in children and patients with syringomyelia. However, PFDD was also associated with a higher rate of overall complications. There was no significant difference in the rate of radiological improvement between the two groups.

Clinical improvement in patients with CM-I following surgery is one of, if not the, most important outcome measure. However, only 13% of case series examined

had usable outcome measures. There is no agreed assessment scale with multiple groups suggesting different scores including Karnofsky, the Chicago outcome score, the Chiari Severity Index, and Klekamp's outcome scoring system, amongst others [10, 11, 23]. It was also not possible to ascertain if, in patients who improved, there was a substantial difference in the amount of their improvement between PFD and PFDD. As there was disparity between the outcome measures reported, all indications that the patient was better were used. In adults, individual comparisons of clinical improvement failed to reach statistical significance. Adults have more symptoms than children, and a longer clinical presentation, which may suggest a poorer prognosis. In adults, there was a trend towards PFDD being better, but further supportive studies are required if this is to reach statistical significance.

Patients with syringomyelia are more likely to improve after PFDD, and this may relate to better reconstruction of the flow dynamics at the craniocervical junction. Radiographic improvements as defined by a reduction in syrinx diameter are reported to appear 10 months after patients experience clinical improvement, and some clinicians see it as an absolute way of defining a successful procedure [2]. The absence of radiographic improvement in any group, may

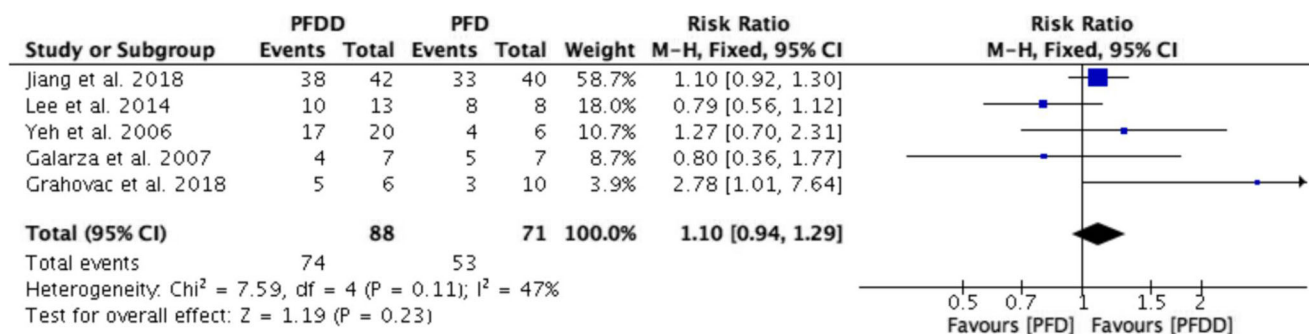


Fig. 8 Radiological improvement—adults

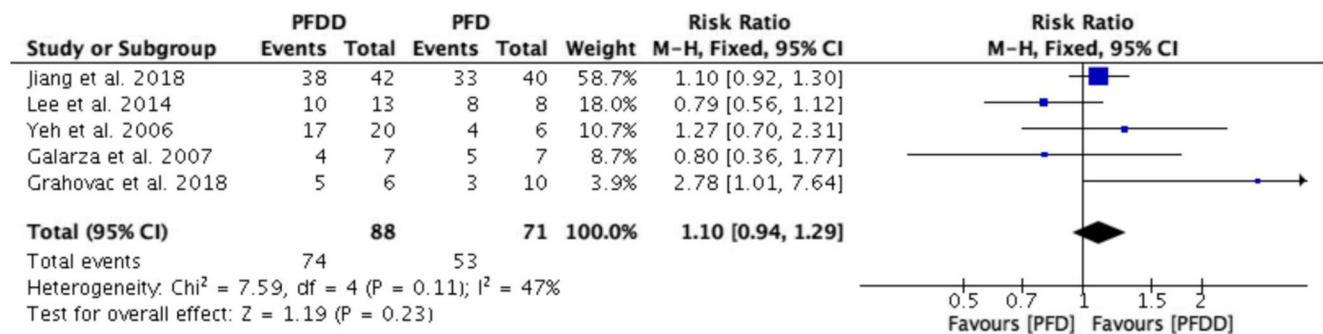


Fig. 9 Radiological improvement—children

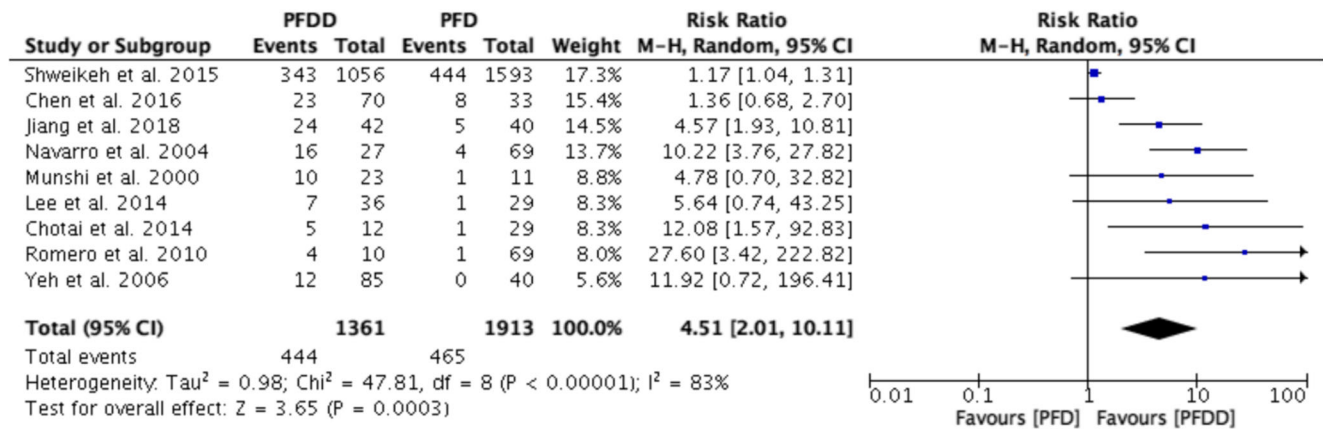


Fig. 10 Overall complications

relate more to the timing of follow-up imaging and the definition of this improvement, rather than being a true reflection of clinical outcome [29].

Published complication rates are higher in patients who have PFDD. A PFDD is more technically challenging than PFD. To reduce complication rates, options include that PFDD only be performed: (1) after failed PFD, (2) in patients with a syrinx, or (3) by surgeons specializing in this area and performing high-volume surgery and/or with low complication rates. In oncology surgery, a large registry study demonstrated a 20% risk reduction in 30-day mortality with doubling of a surgeon's numbers [28]. No

such data currently exists for Chiari surgery, but CSF leak rates of less than 1% are reported in the larger series [14]. Limiting Chiari surgery to high volume, or at least low complication rate, surgeon is surprisingly controversial, but the idea of specialization is supported in other surgical specialties [14].

Graft choice was not examined, and may affect clinical outcomes and complication rates. One study suggested a superiority of autologous pericranium graft in duraplasty, whilst in another series expanded polytetrafluoroethylene grafts were better. A questionnaire of Chiari surgeons suggested autologous

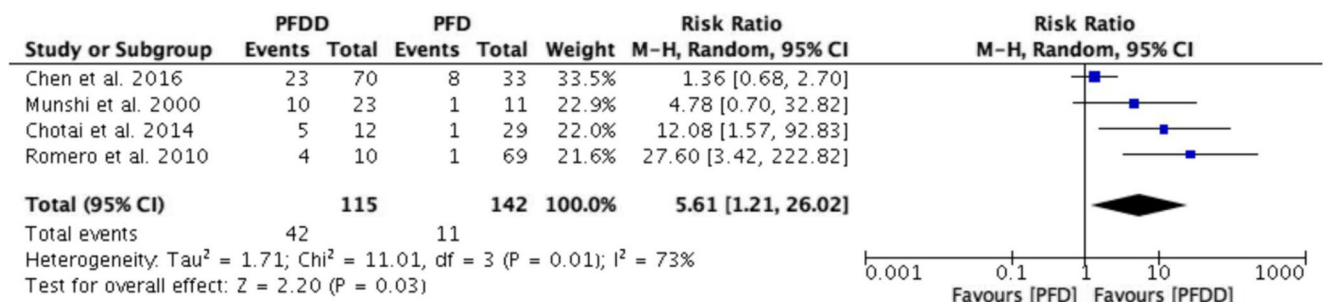


Fig. 11 Overall complications—adults

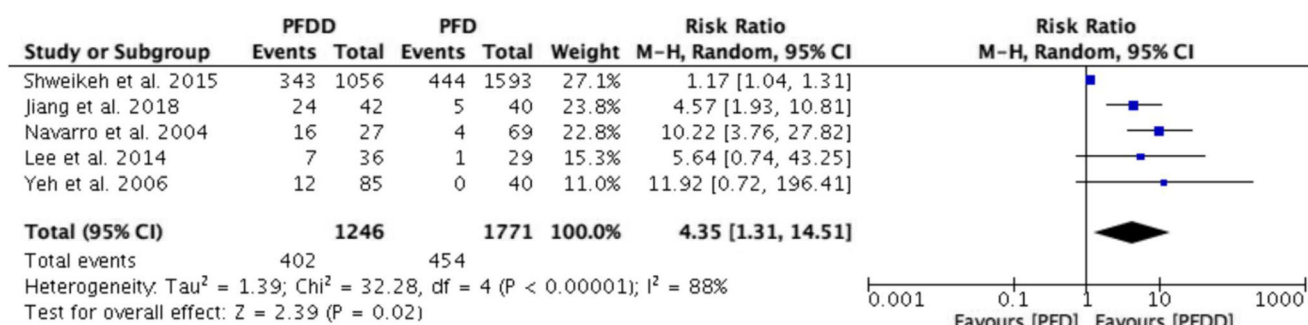


Fig. 12 Overall complications—children

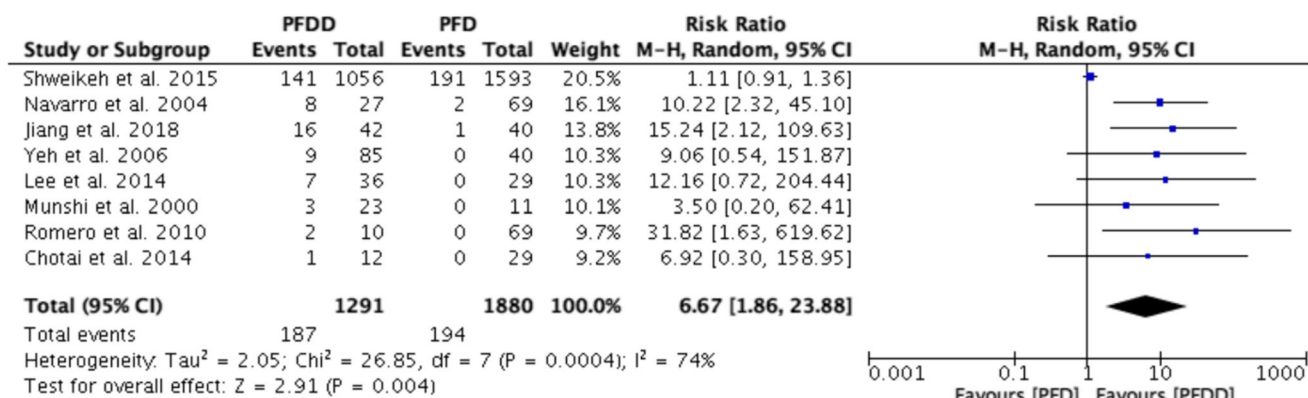


Fig. 13 CSF-related complications

pericranium or bovine pericardium were the most commonly used graft materials [7].

Our study showed no significant difference in reoperation rates between PFD and PFDD. Following the initial operation, a lack of improvement and the recurrence of symptomatology may occur in some patients, hence requiring further operation. The causes of failure range from inadequate dura opening to excessive removal of bony decompression. To minimize the occurrence of these circumstances, Brock et al. suggested the

use of intraoperative ultrasound for CM-I surgery to help determine whether decompression is adequate or if duraplasty is required [4].

Strength and limitations

A meta-analysis allows an assessment and summary of a body of outcome literature. The strength of this study was its straight adherence to PRISMA guidelines, and the use of other

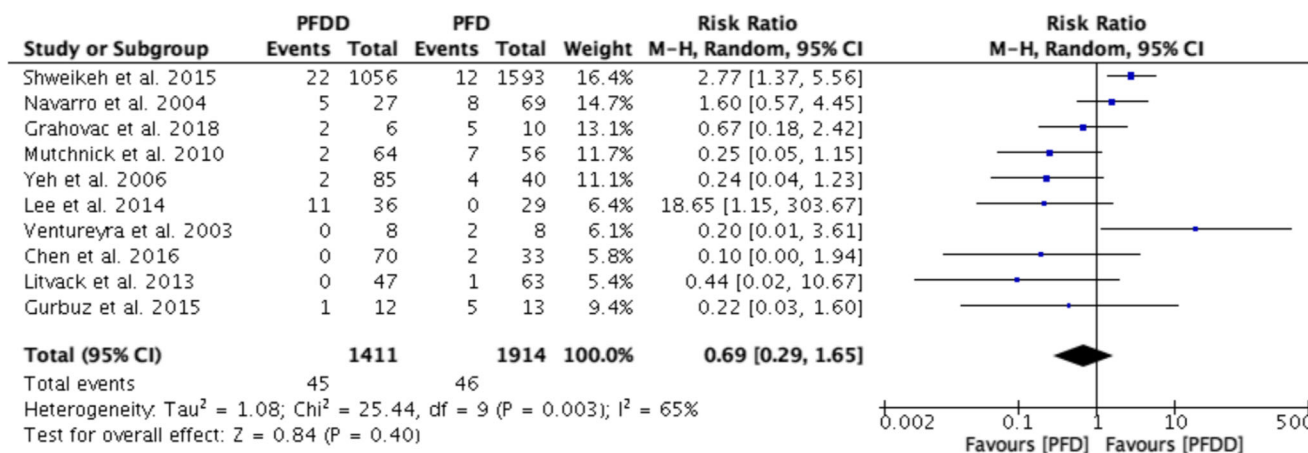


Fig. 14 Reoperations

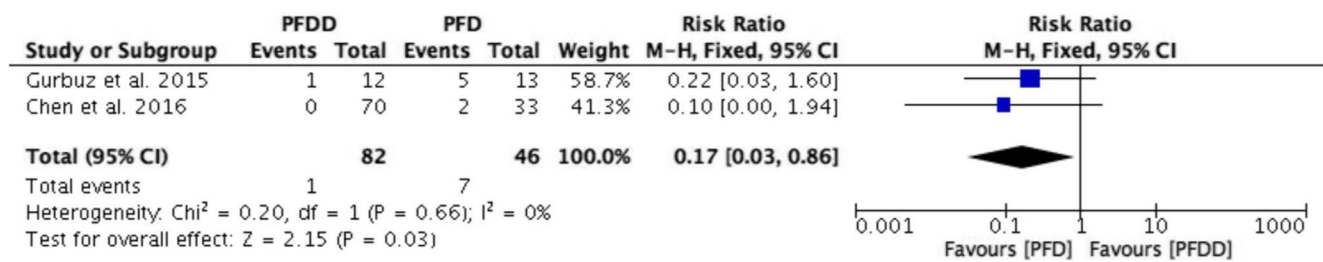


Fig. 15 Reoperations—adults

methods (Newcastle-Ottawa scale, sensitivity analysis) to minimize the effects of heterogeneity between studies on the outcomes presented.

Some limitations of the current meta-analysis must be considered. The series examined were limited to English-language and were mostly unrandomized small cohort studies. There were no standard clinical or radiographic definitions used to define either the Chiari or the outcome. The lack of an agreed standardized outcome measure for Chiari surgery makes comparisons between studies more difficult, and the heterogeneous outcomes reported more difficult to interpret. However, a simple better, same, or worse system provides a simple assessment, which allows comparison. Surgical approaches were based upon the surgeon's training and individual preferences, and not randomized.

PFD includes three types of surgery: a bone only decompression, and in some cases a bony decompression followed by dural opening either with primary closure or leaving the dura open. It was not always clear in the articles reviewed how many of each type was included, so they were combined. This heterogeneity of technique has not been noted before in other systemic reviews. The comparison is therefore between PFDD and a group of operations. There are other technical considerations in Chiari surgery that could also influence the outcome. These include the size of the bone opening, the experience of the surgeon, the amount of blood allowed into the CSF space during the procedure, and the use of tonsillar

reduction, amongst others. A recent review in children failed to find a benefit from tonsillar reduction in their initial series, although no long-term implication of the resection where found [15].

CM-I severity is variable both radiologically and clinically. Such severity is likely to have a major impact on decision-making regarding the choice of approach, and may contribute to the eventual outcome. For instance, patients with tonsillar descent more than 10 mm might do better with more radical surgery. Future analysis based on a standardized grading system may help demonstrate more clear differences in outcome between PFD and PFDD, and with other nuances of surgical technique.

Conclusions

In children with CM-I and in the absence of hydrocephalus and craniocervical region instability, PFDD provides a better clinical outcome, but with higher risk. In adults the risk is higher, and the benefit did not reach significance. In all patients with a syrinx, PFDD appears to provide a better outcome. To help with future outcome assessments in patients with CM-I, standardization of clinical and radiological grading systems is required. A radiological grading system might include the extent of tonsillar descent and degree of impaction.

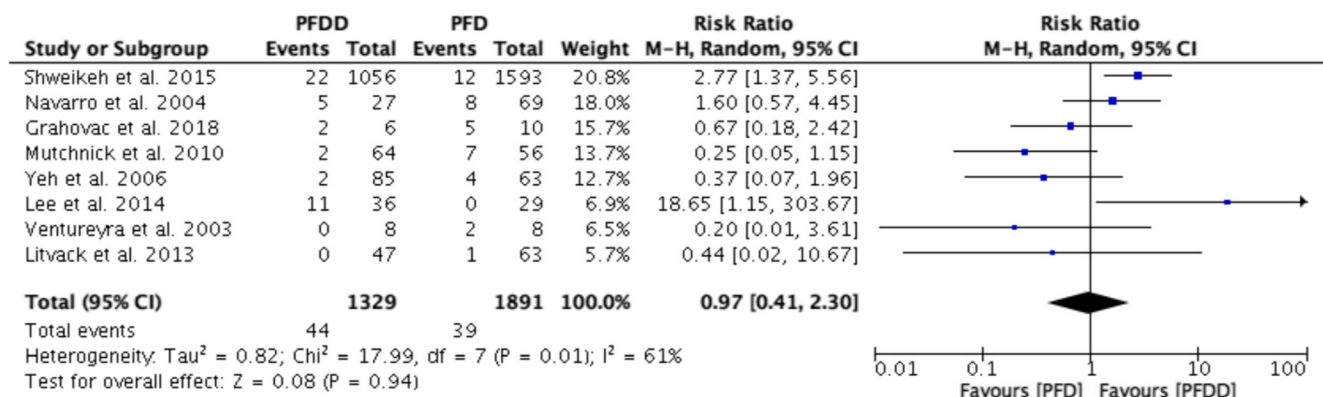


Fig. 16 Reoperations—children

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval For this type of study, formal ethical approval was not required.

Consent For this type of study, formal patient consent was not required.

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