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REVIEW

Posterior fossa decompression with and without duraplasty for the treatment of Chiari malformation type I—a systematic review and meta-analysis

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Abstract The treatment of Chiari malformation type 1 (CM-I) with posterior fossa decompression without (PFD) or with duraplasty (PFDD) is controversial. Our aim is to compare the clinical outcome between the two methods for the treatment of CM-I. In this paper, the authors report a systematic review and meta-analysis of operation time, clinical improvement, and complications of PFD compared with PFDD for the treatment of CM-I. Randomized or non-randomized controlled trials of PFD and PFDD were considered for inclusion. Twelve published reports of eligible studies involving 841participants meet the inclusion criteria. There is significant difference in the operative time [mean difference = −74.63, 95 % CI (−83.02, −66.25), $p < 0.05$] in favor of PFD compared with PFDD. There is significant difference in overall complication rates [mean difference = 0.34, 95 % CI (0.19, 0.60), $p < 0.05$] and rates of CSF leak [mean difference = 0.24, 95 % CI (0.07, 0.78), $p < 0.05$] in favor of PFD groups. However, there is significant difference in the clinical improvement rate in favor of the PFDD group [mean difference = 0.85, 95 % CI (0.73, 0.99), $p < 0.05$]. Although PFDD is related with longer operation time and higher CSF leak rate, it can still be considered as a preferable treatment option for most CM-I patients for its higher improvement rate. More evidence from advanced multi-center studies are needed to provide illumination for the surgical decision making of CM-I.

Keywords Posterior fossa decompression · Duraplasty · Chiari malformation type I · Systematic review · Meta-analysis

Introduction

The Chiari I malformation (CM-I) is the most common condition in the field of craniocervical junction [2, 4]. Surgery is the only treatment available for CM-I; however, multiple methods coexist for this procedure. Posterior fossa decompression with (PFDD) or without duraplasty (PFD) is the surgical treatment of choice [5, 18, 23]. With the trend towards minimally invasive techniques, the duraplasty portion of the procedure has become debatable for the elimination of it can lead to decreased complications. However, PFD is likely to cause higher rates of reoperation as it displays a tendency of inadequate depression [3]. Currently, the consensus on ideal surgical intervention for this condition has not been reached.

This paper will focus on the current literature examining outcomes following PFD compared to PFDD. We report a comprehensive systematic review and meta-analysis to evaluate the clinical advantages of the two methods for the treatment of CM-I.

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Methodology

Search criteria

All full text randomized and non-randomized controlled trials, comparing the clinical outcomes of PFD and PFDD for the patients with CM-I in published studies, were included. Case reports of less than ten subjects, comments, letters, editorials, protocols, guidelines, animal studies, and cadaver articles were excluded.

Search strategy

The Medline, Embase, Cochrane library, Ovid, and CBM databases were searched for English-language articles published from May 1995 to May 2015. Unpublished studies were excluded. Prespecified search terms were "posterior fossa decompression", "duraplasty," and "chiari malformation type I." Titles, abstracts, and subject headings were searched. The reference lists of all included articles and review papers were scrutinized for additional publications.

Search selection

Two reviewers independently assessed the titles and abstracts of each identified citation. The full text of potential articles were ordered and evaluated against the eligibility criteria. Any disagreements were resolved by discussion.

Data extraction

Each reviewer extracted data independently from each included paper. All data was tabulated onto a predefined spreadsheet. All articles were anonymized for author name, institution, journal title, and year of publication to blind reviewers during data extraction, appraisal, and analysis.

Outcome measures

The outcome measures were operation time, complications (neurological complications, cerebrospinal fluid (CSF) leak, wound infection, pseudomeningocele, and aseptic meningitis), clinical improvement, and recurrence rate.

Analysis

The mean differences and 95 % confidence intervals of each outcome were assessed by comparing PFDD and PFD groups, and the statistical heterogeneity was measured by using I^2 statistics. The I^2 test for heterogeneity was used to measure the proportion of total variation in study estimates due to heterogeneity rather than sampling error. If significant heterogeneity was found among studies based on interpretation of the I^2 test, a random effects model was applied. If no significant heterogeneity among studies was found, a fixed-effects model was applied. After this, the meta-analysis was carried out by using REVMAN software (version 5.0 for Windows. Copenhagen: The Nordic Cochrane Centre, the Cochrane Collaboration, 2008). p Values of less than 0.05 were considered statistically significant. We attempted to contact the original authors and inquire insufficient data of them. We also approached missing standard deviations for changes from the baseline by referring to Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0 (16.1.3.2).

Results

Search strategy

We found 864 potentially eligible articles of which 12 studies were included [7–10, 12, 13, 15, 16, 19–21, 25, 31] (Fig. 1). A total of 841 patients were included, 469 of which received PFD and the other 175 adopted PFDD. The follow-up periods were more than 6 months (Table 1).

Outcome measure

Mean operative time

There studies [15, 16, 21] recorded the mean operative time. A meta-analysis showed there is significant difference in the operative time [mean difference = −74.63, 95 % CI (−83.02, −66.25), $p < 0.05$] (Fig. 2).

Complications

Twelve studies [7–10, 12, 13, 15, 16, 19–21, 25, 31] recorded the complications including neurological complications, CSF leak, wound infection, pseudomeningocele, and aseptic meningitis. There is significant difference in the complications between the two groups in favor of PFD [mean difference = 0.34, 95 % CI (0.19, 0.60), $p < 0.05$]. There is significant difference in the rate of CSF leak between the two groups [mean difference = 0.24, 95 % CI (0.07, 0.78), $p < 0.05$]. On the other hand, there is no significant difference between the two groups in the occurrence rate of neurological complications [mean difference = 0.25, 95 % CI (0.06, 1.10), $p > 0.05$], pseudomeningocele [mean difference = 0.25, 95 % CI (0.05, 1.21), $p > 0.05$], and aseptic meningitis [mean difference = 0.26, 95 % CI (0.05, 1.22), $p > 0.05$] and in the wound infection rate [mean difference = 0.86, 95 % CI (0.28, 2.68), $p > 0.05$] (Figs. 3 and 4).

Fig. 1 A QUORUM chart

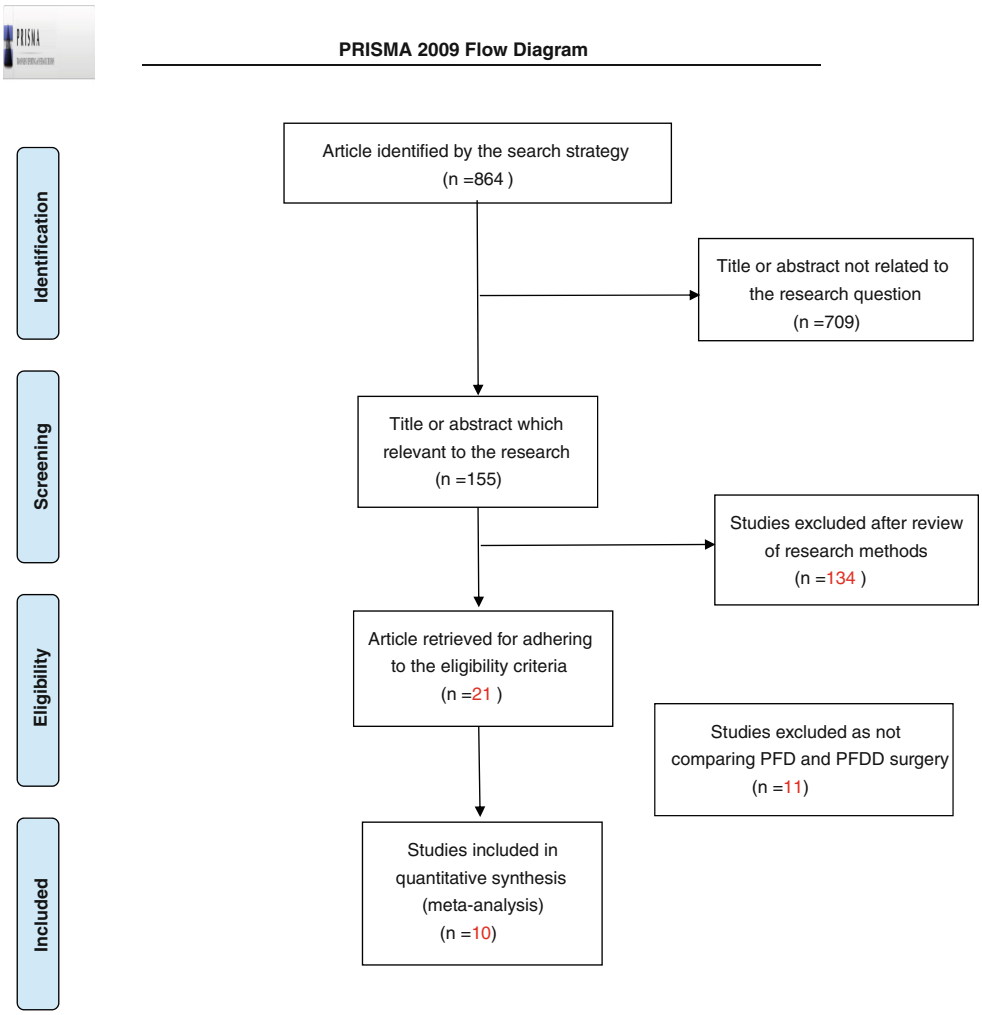


Table 1 General condition of the 12 studies included in the meta-analysis

Paper	Operation		Mean age		Gender (M/F)		Follow-up		Outcomes
	PFD	PFDD	PFD	PFDD	PFD	PFDD	PFD	PFDD	
Chotai 2014	29	12	33.8 years		8/34		1.2 years		Clinical improvement, complications
Erdogan 2010	12	15	31.58 years	25.86 years	9/3	15/0	NA		Clinical improvement, complications
Galarza 2007	20	21	10 years		NA		1.8 years		Clinical and imaging improvement, complications
Gurbuz 2015	18	21	36 years		13/26		3.6 years		Complications
Lee 2014	29	36	8.9 years	9.9 years	15/14	16/20	2y	1.9 years	OR time, imaging improvement, complications
Limonadi 2004	12	12	7.6 years	10.8 years	5/7	6/6	1.3y		OR time, complications
MutchnIck 2010	56	64	11.1 years		58/63		0.5 years		OR time, complications
Yilmaz 2011	24	58	38.9 years	31 years	36/46		0.8y		Clinical and imaging improvement, complications
Romero 2010	6	10	40.6 years		6/10		1.4 years		Clinical improvement, complications
McGrit 2007	116	140	29 ± 15 months		121/135		NA		Complications
Munchi 2000	11	23	32.5 years		11/23		1–6 months		Clinical improvement, complications
Hayhurst 2008	84	12	33.0 years		35/61		6 months–9 years		Clinical improvement, complications

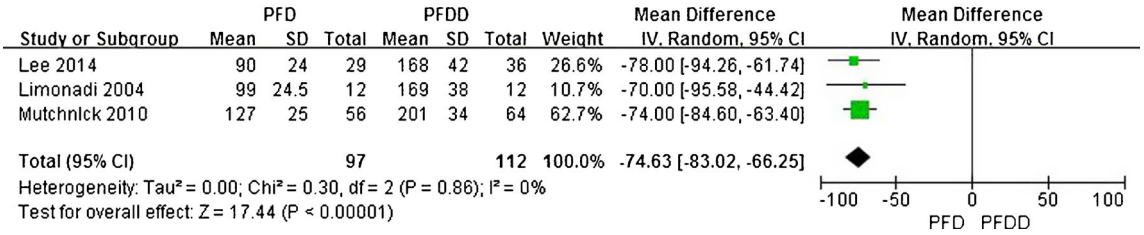
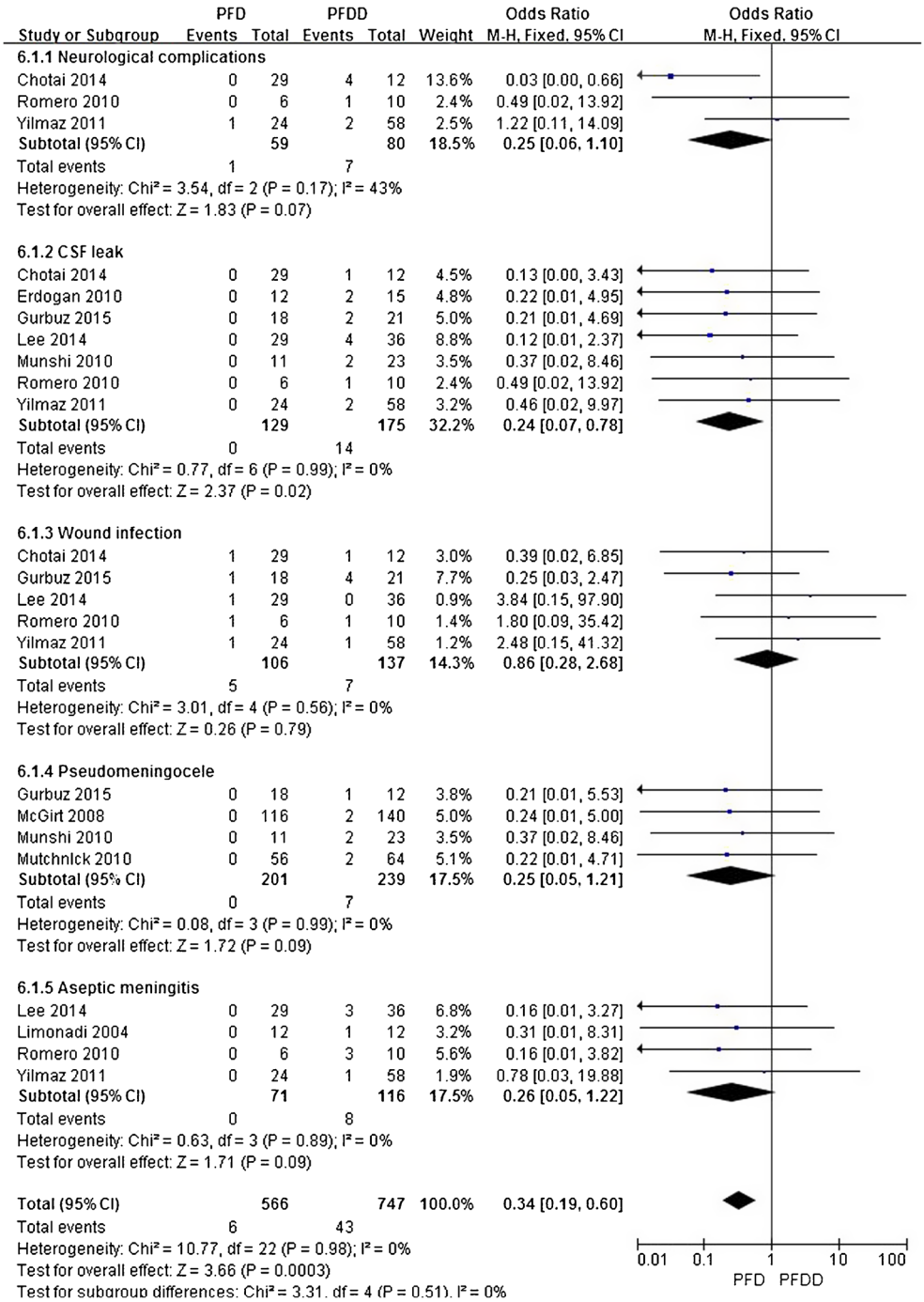


Fig. 2 A meta-analysis showed there is significant difference of the operative time [mean difference = −74.63, 95 % CI (−83.02, −66.25), $p < 0.05$]

Fig. 3 There is significant difference in the complications between the two groups in favor of PFD [mean difference = 0.41, 95 % CI (0.30, 0.57), $p < 0.05$]. Specifically speaking, the occurrence rates of neurological complications [mean difference = 0.27, 95 % CI (0.07, 0.96), $p < 0.05$], CSF leak [mean difference = 0.16, 95 % CI (0.05, 0.51), $p < 0.05$], pseudomeningocele [mean difference = 0.10, 95 % CI (0.02, 0.53), $p < 0.05$], and aseptic meningitis [mean difference = 0.21, 95 % CI (0.05, 0.88), $p < 0.05$] are all significantly higher in the PFDD group compared to the PFD group. However, there is no significant difference in the wound infection rate between the two groups [mean difference = 0.86, 95 % CI (0.28, 2.68), $p > 0.05$]



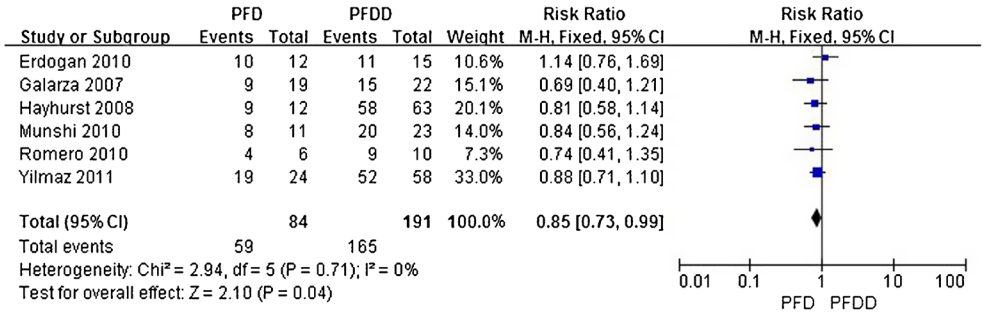


Fig. 4 There is no difference in the clinical improvement rate between the two groups [mean difference = 0.55, 95 % CI (0.27, 1.10), $p > 0.05$]. Subgroup analysis shows that improvement rate is not significant both in children [mean difference = 0.49, 95 % CI (0.18, 1.35), $p > 0.05$] and adults [mean difference = 0.55, 95 % CI (0.23, 1.60), $p > 0.05$]

Clinical improvement

Six studies [9, 10, 13, 16, 21, 31] recorded the clinical improvement rate. There is significant difference in the clinical improvement rate between the two groups in favor of the PFDD group [mean difference = 0.85, 95 % CI (0.73, 0.99), $p < 0.05$].

Critical appraisal

The population and study eligibility criteria are accurately defined, insuring comparable parameters are balanced and efficient in the study groups. Related research questions can be clearly addressed by appropriate selection and definition of the outcome measures. However, methodological limitations still exist in the literature (Table 2). Several factors may bias the findings of the current study. First, bias can be introduced in a retrospective review that does not have randomized, prospectively matched groups. Second, because both the practitioners and evaluators are surgeons, the design of the project cannot be absolutely blinded to which surgery was operated and clinical improvements such as numbness and tingling are hardly objective. Third, Some studies may reserve bony decompressions for mild cases or patients without syringomyelia and PFDD for the more severe cases which could incur bias. In both PFD and PFDD groups, the bony decompression range may also influence our results. Other confounding factors could be the operative decisions and techniques of the different surgeons. Some studies' follow-up periods are quite short, and it may also affect the results. Multiple alternative factors, such as the implementation of tonsillar resection, the adoption of intraoperative ultrasound, and the different material used for duraplasty, could affect the outcome. This, therefore, permitted bias.

Discussion

The CM-I constitutes a group of congenital or acquired etiology that has descent of the cerebellar tonsils into the cervical

Table 2 Risk of bias assessment of included studies

Paper	Adequate sequence generation?	Allocation concealment used	Blinding	Interventions clearly defined	Outcome measures clearly defined	Outcome measures appropriate	Appropriate follow-up duration
Chotai 2014	Yes	No	No	Yes	Yes	Yes	Yes
Erdogan 2010	Yes	No	No	Yes	Yes	Yes	Unclear
Galarza 2007	Yes	No	No	Yes	Yes	Yes	Yes
Gurbuz 2014	Yes	No	No	Yes	Yes	Yes	Yes
Lee 2014	Yes	No	No	Yes	Yes	Yes	Yes
Limonadi 2004	Yes	No	No	Yes	Yes	Yes	Yes
MutchnIck 2010	Yes	No	No	Yes	Yes	Yes	Yes
Yilmaz 2010	Yes	No	No	Yes	Yes	Yes	Yes
Romero2010	Yes	No	No	Yes	Yes	Yes	Yes
Munchi 2000	Yes	No	No	Yes	Yes	Yes	Yes
Hayhurst 2008	Yes	No	No	Yes	Yes	Yes	Yes
McGrit 2007	Yes	No	No	Yes	Yes	Yes	Yes

spinal canal. The surgical indications of CM-I were usually headache (or tussive headache); neck, arm, or back pain; swallowing difficulties; drop attacks; or upper extremity numbness or tingling. The presence of a syrinx was also an indication for surgery when it occurred in the presence of the above symptoms [4, 5, 11, 20].

For these patients and neurosurgeons, the clinical improvement is the most priority for the surgical decision making. Generally, the postoperation symptomatic improvement has been reported in the range of 61.5–93 % from different studies [5, 6, 8, 16, 17]. Some studies indicated that PFDD is better at least for patients with syringomyelia and some report clinical results as comparable [21, 30]. According to our results, PFDD can achieve more satisfactory outcomes than PDF can in the clinical improvement between the two groups.

Some studies also reported the imaging improvement which is not as good as the clinical improvement [10, 12, 15, 31]. Though the compression has been relieved by surgical decompression, arachnoid scarring prevents the nervous structures to regain its normal position [10, 24]. However, the minimal enlargement of the subarachnoid spaces at the craniocervical junction resulting from bone decompression was sufficient to relieve the impact on the nervous structures and to improve CSF circulation. There is also no significant correlation between the reduction in syrinx size on MRI and the degree of clinical improvement [8, 10, 16, 29, 30]. Although clinical symptoms do not correlate to the presence or size of preoperative syringomyelia, there can be no doubt that a permanent postoperative reduction of syrinx size is an indicator of a sufficient decompression. On the contrary, the lower numbers for postoperative syrinx reductions in the bony decompression group must be seen as a prognostic indicator for worse long-term results. It implies that neurosurgeons should focus on both clinical outcome and radiological findings.

To better evaluate the effectiveness of PFD and PFDD in the management of CM-I, many researchers reported the re-operation rates [7, 9, 10, 15, 21, 22]. Once it occurs, the patients will suffer from tremendous pain and heavy economic burden. We hold the opinion that the re-operation may be caused by two main reasons: the persistence symptoms or the severe complications. Due to insufficient data, the different reasons of re-operation cannot be meta-analyzed in this paper. However, according to some research and our experience, more second surgeries had been done because of the recurrent syndromes such as nausea and vomiting rather than severe complications. Many studies observed a trend for lower recurrence rates in the PFDD group; whether this trend reaches the level of significance remains to be tested. For the patients who have recurrent clinical symptoms after PFD, a second PFDD can achieve better effect [1, 8, 15, 21, 26, 27].

For the other important aspect, PFD is associated with lesser rate of overall complications compared with PFDD especially in the CSF leak complication according to our results which reduces the re-operation to a great degree. The CSF-related complications include postoperative CSF leak, pseudomeningocele formation, meningitis, and scarring of the arachnoid leading to obstruction of CSF dynamics and foreign-body reaction [1, 3]. Most complications are related to CSF exposure to blood and muscle cellular debris as well as the use of dural graft. Thus, extradural decompression without duraplasty will be associated with lesser complication rates. Like previous studies [16, 23], we also found that the PFD group had shorter operative time than did the PFDD group. Besides, many researches have reported that PFD leads to shorter hospital stay lengths and less hospitalization costs [8, 17, 27].

CM-I is often associated with other medical conditions such as syringomyelia. Recent studies have reported an incidence rate as high as 70–80 % for syringomyelia [10, 28–30]. The range of the decrease rate of syringomyelia is wide (55–100 %) according to previous reports [8–10, 12, 16, 20, 26, 30]. Though not statistically significant, some findings also show higher rates of syrinx improvement in patients undergoing PFDD [23, 28–30]. Despite these findings, recently, a large sample study by Shweikeh [27] shows that CM-I patients with syringomyelia or hydromyelia were more often treated with PFD rather than PFDD. It is possible that the presence of syringomyelia does not currently influence treatment decisions.

Above all, for the criteria of the surgical decision making, we believe that for those with rapidly progressive symptoms or severe neurological deficits, previous research advises PFDD as the first option. While for patients with mild symptoms and no syringomyelia, PFD could be the first choice. If symptoms fail to improve, PFDD should be considered. According to our results, PFDD is better with improving rates but with more surgical trauma and higher rate of CSF leak. So, which is the preferable choice for those patients? As for the decompression surgery, our aim is to restore normal CSF dynamics at the level of the craniocervical junction. Thus, intraoperative ultrasonography may be a useful tool to aid the surgeon in deciding whether to adopt PFD or PFDD and even tonsillar shrinkage. Recently, more and more studies demonstrate that intraoperative ultrasonography can effectively guide the surgical decision making [14, 19, 32].

Conclusion

Although PFDD is related with longer operation time and higher CSF leak rate, it can still be considered as a preferable treatment option for most CM-I patients for its higher improvement rate. And PFDD tends to be worthy of consideration under the circumstance of failed PFD. More evidence from advanced multi-center studies are needed to provide illumination for the surgical decision making of CM-I.

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Compliance with ethical standards

Competing interests The authors declare that they have no competing interests.

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Comments

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The treatment of Chiari type I malformation continues to raise interest and debate. A well-known issue concerns the management of asymptomatic/poorly symptomatic subjects. A further and still disputed one concerns the best surgical operation. Actually, many kind of surgical treatments are adopted in different centers, ranging from the bony decompression of the posterior cranial fossa alone (craniectomy with or without C1 laminectomy) to the coagulation of tonsils passing through the expanding decompressive craniotomy, the dural delamination, and the duraplasty. All these surgical approaches seem to ensure good clinical results with some differences as far as the radiological outcome and the complications are concerned. On these grounds, an updated systematic review and meta-analysis of the literature was needed.

The review proposed by Xu and coworkers is focused on the "dilemma" between posterior fossa decompression alone (PFD) or with duraplasty (PFDD). As expected, PFD showed lower operating times and lower rates of complications compared with PFDD. However, PFDD showed better clinical outcome and lower risk of recurrence. Similar results were collected by Durham and Fjeld-Olenec in a previous meta-analysis on pediatric patients [8]. The confirmation provided by Xu et al. in a larger and mixed population (children and adults), therefore, would allow the neurosurgeons to definitely identify the PFDD as more effective than PFD, though more risky. Nevertheless, as stressed by the authors, several biases burden the study, as the different criteria for indication and evaluation of the results adopted by the authors of the analyzed papers, the use of retrospective series, the extent of bone decompression, the length of follow-up, or the materials used for the duraplasty. In addition, the inability to achieve a systematic correlation between clinical and radiological findings deprives this meta-analysis of information on the trend of the CSF dynamics in the posterior fossa (cisterna magna re-expansion) and the spinal cord (decrease of syringomyelia). This reinforces the need for multicenter randomized trials.

The goal of the surgical treatment of Chiari type I is to restore the CSF spaces at the craniocervical junction. In spite of the obtained results, the authors do not solve the dilemma about the best solution between PFD and PFDD. They propose intraoperative ultrasounds as the method to evaluate the restoration of the cisterna magna and, consequently, the need for a duraplasty. Such a strategy is already used by many authors (including the author of the present comment) and gives reasons of some results found in the literature and some possible biases. Indeed, intraoperative MRI, utilized for the same purpose in a recent prospective series, demonstrated an improvement of the CSF flow with the prone position alone so good that it was not significantly enhanced after PFD (Bond AE et al., J Neurosurg 122: 1068–1075, 2015). This observation, unfortunately, offers a further intraoperative bias. For these reasons, the surgical management of Chiari I should continue to be tailored on the single patient based on preoperative clinical and radiological criteria (PFD for poorly symptomatic patients without syringomyelia, PFDD for clearly symptomatic ones with syringomyelia) other than on intraoperative radiological criteria. Similarly, children should be differentiated from adults. Actually, pediatric patients do not infrequently show a moderate tonsillar herniation that is hard to be correlated with the clinical picture and that is not associated with syringomyelia (or is associated with thin hydromyelia). In these instances, also due to the residual potential of growth of the posterior fossa and the increased risk of CSF leakage in children, PFD alone should be considered.

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The authors of this paper propose to leave the dura open after decompression of the foramen magnum for patients with Chiari I malformation in order to avoid problems with dura grafts and formation of arachnoid adhesions between dura or duragraft and underlying cerebellar and spinal cord tissue. This is a concept originally used by Gardner in his paper from 1965. Bernard Williams adopted this strategy fearing reobstruction of CSF pathways by arachnoiditis when dura grafts are used. He was convinced that duraplasties should be avoided whenever creating a sustained CSF passage was part of the surgical strategy. I had many personal discussions with him on this subject without ever agreeing on this issue. While working in Hannover, a number of patients were actually operated leaving the dura open. Their results, however, were considerably worse compared to patients operated with duraplasties, so this technique was quickly abandoned:

1. Leaving the dura open allows breakdown products of blood or muscle proteins to contaminate the subarachnoid space. This causes a severe arachnoiditis. If such areas are reopened surgically a few months or years later, the entire area usually appears covered by a thick, whitish membrane. It is no longer possible to identify any blood vessels on the cord surface or caudal cranial nerves lateral of the cervical cord and medulla oblongata. If the foramen of Magendie is closed by this membrane, any attempt of opening it is extremely dangerous for lack of any anatomical landmarks. When this technique had been used in the spinal canal, the CSF passage was regularly found to be obstructed due to this arachnoiditis upon reopening.

2. Leaving the dura open puts patients at risk to develop superficial siderosis, which is a potentially life-threatening complication related to repeated contaminations of CSF with blood. Bernard Williams operated on a woman with Chiari I malformation in Hannover during an instructional course in 1990 leaving the dura open. The patient did well postoperatively for about 1 year when she started to demonstrate signs of this disease causing severe gait and hearing problems.

3. With time, bulging of neck muscles may lead to a progressive decrease of the subarachnoid space at the foramen magnum even to the point of complete obstruction of CSF flow. Although I have seen such cases presented in scientific meetings by British neurosurgeons, who widely adopted Williams' technique in the 1980s and 1990s, this late complication has not been published to my knowledge.

Therefore, I cannot agree with the authors of this paper and strongly advise against the technique of leaving the dura open for patients with Chiari decompressions or other pathologies.

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In this interesting paper from Xu et al., the authors perform a meta-analyis of the last two decades literature in order to clarify if posterior fossa decompression with (PFDD) and without (PFD) duroplasty shows differences in terms of outcomes.

The quest for the perfect intervention on Chiari I malformation (CMI) has led many authors to try many different approaches. By looking at the last decades, surgery for CMI has become more minimally invasive with the aim to reducing complications. In fact, intraarachnoid manipulation, although performed with success by several authors, intrinsically exposes the patient to risks. Moreover, adding tonsillar and arachnoid manipulation does not seem to bring further improvement to outcomes. Proposing an osseous decompression without duroplasty has become the natural consequence of chasing the most atraumatic surgery. Unfortunately, as pointed out in this meta-analysis, leaving the dura mater intact can potentially augment the number of redo surgery due to an insufficient

decompression and eventually scarce symptom relief. We routinely perform PFD with duroplasty in all subjects. This said, PFD without duroplasty is still the preferred treatment in children and adolescents since the CSF dynamics follows completely different rules in young patients leading to higher numbers of external fistulas. In our opinion, this strategy is also preferable in those rare elderly patients where the aim is to guarantee relief of symptoms while avoiding any possible complications and reducing operative times.

One aspect that should be regarded with interest is that, despite different technical nuances, recent literature demonstrates that a certain percentage of patients does not benefit from decompression. It is difficult sometimes to predict clinical result, but today, it is clear that one of the first causes of unsatisfying results is an incomplete diagnosis. This implies not only to mistake a cerebellar tonsils' ecotopia with CMI but also to neglect many other structural alterations (craniocervical-associated malformations, instability, cerebrospinal fluid's dynamic alterations) which require sometimes different or multiple treatments.