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REVIEW

Radiological outcome after surgical treatment of syringomyelia-Chiari I complex in adults: a systematic review and meta-analysis

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Abstract

Foramen magnum decompression (FMD) is widely accepted as the standard treatment for syringomyelia associated with Chiari type I malformation (CMI). Despite extensive clinical investigations, relevant surgical details are still matter of debate. The authors performed a systematic review and meta-analysis of the literature examining the radiological outcome of syringomyelia in adult patients with CMI after different surgical strategies. PRISMA guidelines were followed. A systematic search of three databases was performed for studies published between 1990 and 2018. Our systematic review included 13 studies with a total of 276 patients with CMI associated with syringomyelia. Overall, the rate of post-operative radiological improvement at last follow-up was 81.1% (95% CI 73.3–88.9%; $p < 0.001$; $I^2 = 71.4\%$). The rate of post-operative syrinx shrinkage did not differ significantly among both groups of decompression with the extra-arachnoidal technique and arachnoid dissection (90%, 95% CI 85.1–94.8%, $I^2 = 0\%$ vs 79.8%, 95% CI 61.7–98%, $I^2 = 85.5\%$). A lower rate of post-operative radiological syrinx shrinkage was observed after decompression with splitting of the outer layer of the dura (55.6% 95% CI 40.5–70.8%, $I^2 = 0\%$). CSF-related complications and infections were similar among the different groups. Our meta-analysis found that FMD with the extra-arachnoidal technique and arachnoid dissection provides similar results in terms of post-operative shrinkage of syringomyelia. Patients undergoing decompression with splitting of the dura presented the lower rate of syrinx reduction. These data should be considered when choosing the surgical approach in adult patients with CMI associated with syringomyelia.

Keywords Chiari I malformation · Syringomyelia · Surgery · Foramen magnum decompression · Arachnoid exploration · Meta-analysis

Introduction

Surgical procedures for syringomyelia associated with Chiari type I malformation (CMI) are mainly directed toward decompressing cerebellar tonsils at the foramen magnum and restoring CSF flow dynamics to normal. Despite extensive clinical investigations, considerable controversy about the technical details of the operation is still present [25, 37]. The most common technical modalities described in the literature include the following: (1) foramen magnum decompression (FMD) with incision of the external layer of the dura [16]; (2) FMD, dural opening, arachnoid preservation with or without duraplasty [27]; (3) FMD, dural opening, arachnoid dissection with or without coagulation or resection of cerebellar tonsils and duraplasty [17]. However, most studies lack enough patients to allow sound conclusions on how patients with syringomyelia-CMI complex should be operatively treated. In particular, the question about whether the arachnoid should be dissected after the osteo-dural decompression and the efficacy of arachnoid dissection on the post-operative shrinkage of syringomyelia has not been fully elucidated. Improved understanding of the treatment-related radiological outcome after foramen magnum decompression can help

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practitioners in the selection of the surgical procedure for syringomyelia associated with CMI.

The aim of our meta-analysis was to examine and compare the radiological outcome of syringomyelia in patients with CMI after different surgical strategies.

Methods

Literature search

A comprehensive literature search of PubMed, Ovid MEDLINE, and Ovid EMBASE was conducted for studies published from January, 1990 to May, 2018. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-analysis [23]) guidelines were followed. The key words and the detailed search strategy are reported in Table 1.

The inclusion criteria were the following: (1) studies reporting surgical series of adult patients (> 18 years old) with syringomyelia associated with CMI; (2) studies reporting series of patients treated with FMD with or without dural opening and with or without arachnoid dissection; (3) studies detailing post-operative complications and reporting radiological outcome of syringomyelia. Exclusion criteria were the following: (1) studies reporting series with less than 10 patients; (2) studies reporting patients with craniovertebral junction (CVJ) malformations or hydrocephalus; (3) review articles; (4) studies published in languages other than English. In cases of overlapping patient populations, only the series with the largest number of patients or most detailed data were included.

Two independent readers screened articles (F.C. and Y.A.) in their entirety to determine eligibility for inclusion. A third author solved discrepancies (D.D.C.).

Data collection

From each study, we extracted the following: (1) demographic data; (2) number of patients with CMI associated with syringomyelia; (3) preoperative clinical and radiological data; (4) post-operative complications (pseudomeningocele, CSF leakage, aseptic meningitis, and infection). Preoperative clinical symptoms were classified as follows: pain-related (tussive sub-occipital headache, non-tussive headache, neck pain), spinal cord-related (motor signs including paresis/weakness of at least one limb, spasticity, muscle atrophy, sensory symptoms including hypoesthesias, dysesthesias, paresthesias, peripheral nerve neuralgias), brainstem-related (including dizziness, diplopia, nystagmus), hiccup cerebellar-related (ataxia, gait impairment, vertigo, dysarthria), and lower cranial nerve-related (LCN). For each series, we collected site and extent of syrinx.

FMD was sub-grouped as follows: (1) FMD with splitting of the dura (opening of the outer layer only) (FMDds); (2) FMD with dural opening, arachnoid preservation with or without duraplasty (FMDdo); (3) FMD with dural opening, arachnoid exploration, and duraplasty (FMDae) (Fig. 1a–c). In the last sub-group, different surgical maneuvers were reported after arachnoid dissection (such as lysis of arachnoid

Table 1 Search syntax

PubMed search accessed June 2018 (295 articles)	SCOPUS search accessed on June 2018 (192 articles)	EMBASE search accessed June 2018 (194 articles)
((((((((Chiari I malformation[MeSH Terms]) OR arnold chiari syndrome[MeSH Terms]) OR syringomyelia[MeSH Terms])) AND (((surgery[MeSH Subheading]) OR surgical treatment[Title/Abstract]) OR surgical management[Title/Abstract])) AND (((((posterior fossa decompression[Title/Abstract]) OR suboccipital craniectomy[Title/Abstract]) AND subdural decompression[Title/Abstract]) OR duraplasty[Title/Abstract]) OR subarachnoid[Title/Abstract])) AND (“1990/01/01”[Date - Publication]: “2018/05/01”[Date - Publication]))) AND English[Language]	(TITLE-ABS-KEY (chiari AND i AND malformation OR syringomyelia) AND TITLE-ABS-KEY (surgery) AND TITLE-ABS-KEY (sub-dural OR duraplasty OR sub-arachnoid)) AND DOCTYPE (ar) AND PUBYEAR > 1989	(“arnold chiari malformation”/exp/mj OR “syringomyelia”/exp/mj) AND “surgery”:lnk AND “posterior fossa decompression”:ab,ti AND [1990–2018]/py

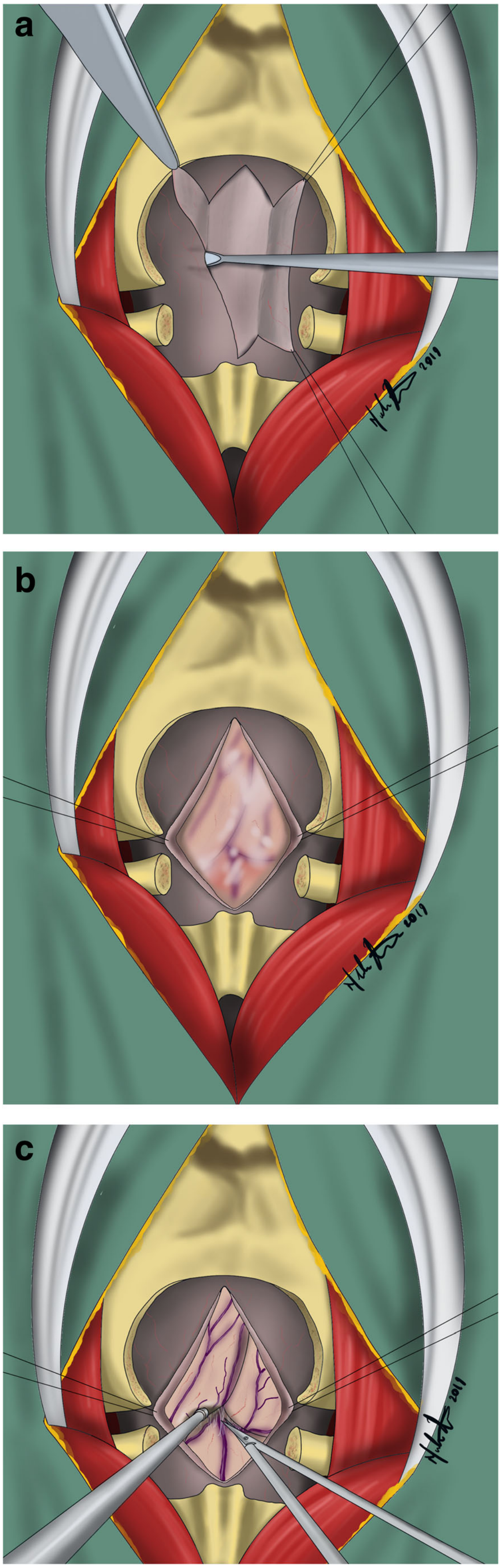

Fig. 1 Illustrations depicting the three modifications of foramen magnum decompression (FMD) investigated in this study. **a** FMD with splitting of the dura (opening of the outer layer). **b** FMD with durotomy (dural opening and arachnoid preservation) with or without duraplasty. **c** FMD with durotomy, arachnoid exploration/dissection, and duraplasty

bands/veils, cerebellar tonsil shrinkage, and plugging of the obex). A sub-analysis of this population was beyond the scope of our meta-analysis, and it was not reported.

Radiological outcome was assessed at the last MRI follow-up and dichotomized as follows: better/resolved and unchanged/worse. Post-operative complications were dichotomized as follows: CSF-related (pseudomeningocele, CSF leak) and inflammatory/infection-related (aseptic meningitis, wound infections, meningitis).

Outcome measures

The primary objective of this meta-analysis was to assess the rate of post-operative radiological improvement of syringomyelia after FMD and to compare the radiological outcome according to the surgical approach used. The secondary objective was to evaluate the impact of potential confounders on the analyzed outcome. The analysis was performed for the following variables: rate of post-operative CSF-related complications, rate of post-operative infections, median follow-up (radiological or clinical), rate of patients with radiological evidence of tonsils below C1, rate of patients with syringomyelia limited to the cervical spine, mean duration of symptoms before surgery. In addition, the analysis of post-operative complications for each sub-group was conducted.

Quality scoring

A modified version of the Newcastle-Ottawa Scale (NOS) [38] was used for the quality assessment of the included studies. The details are reported in Table 2. Two authors performed the quality assessment independently (F.C. and Y.A.) and a third author solved discrepancies (D.D.C.). Studies rated with a NOS ≥ 6 were considered as "high quality."

Statistical analysis

The Wald method was used to calculate confidence intervals for event rates. A two-tailed *t* test was used for continuous data and Fisher's exact test for categorical variables. In order to assess the heterogeneity of the data, the Higgins index (I^2) [15] was used, in which $I^2 > 50\%$ suggests substantial heterogeneity. DerSimonian and Laird random effects model was subsequently applied [8]. The graphical representation of the meta-analysis was performed by forest plot. To verify the consistency of meta-analysis, the influence of each individual study was assessed by the sensitivity analysis ("leave-one-

Table 2 Summary of the studies included in the systematic review, accounted for FMD surgical technique

	Year	Design	*N* patients	M/F	Age (mean)	Syrinx	Median FU (months)
FMDds							
Chauvet et al.[7]	2009	R	10	2\8	34.3	5	12
Kotil et al.[18]	2009	P	10	4\6	44.4	10	25
Ono et al.[26]	2010	P	20	4\16	49.1	20	–
Romero et al.[32]	2010	R	6	3\3	40.7	4	–
FMDdo							
Perrini et al.[27]	2007	R	24	9\15	40	24	44
Rahman et al.[30]	2017	R	11	7\4	29.4	11	36
Rehman et al.[31]	2015	P	21	8\13	–	21	–
Romero et al.[32]	2010	R	10	3\7	40.5	5	–
Spena et al.[33]	2010	R	36	17\19	40.4	36	40
Yilmaz et al.[43]	2011	R	58	–	38.9	45	–
FMDae							
Fischer et al.[12]	1995	P	10[a]	–	49	9	25.5
Pillay et al.[28]	1991	P	31[c]	–	–	17	–
Prat et al.[29]	2009	P	12[b]	3\9	43.6	12	12
Thakar et al.[35]	2018	R	57	30\27	38.3	57	–

Ae arachnoid exploration/dissection, *ds* dura splitting, *do* dura opening, *FMD* foramen magnum decompression, *FU* Follow-up, *P* prospective study, *R* retrospective study

[a] 9 patients excluded; [b] 1 patients < 18 years old excluded; [c] 4 patients excluded due to other neurosurgical procedures; [d] 3 patients excluded due to other neurosurgical procedures

out" approach) [42], and subsequently funnel plot followed by Egger's linear regression test were analyzed [10, 11]. Random effects meta-regression [2] was used to assess significant association between the effect size and the following moderators: surgical technique (FMDds, FMDdo, FMDae), patient's mean age, CSF-related complications, post-operative infections, median follow-up, and study publication year. Differences were considered significant at $p < 0.05$. Statistical analyses were performed with SPSS version 23 (SPSS Inc. SPSS® Chicago, IL, USA), with ProMeta version3 (Internovi, Cesena, Italy) and OpenMeta[Analyst] (http://www.cebm. brown.edu/openmeta/).

Results

Literature review

A total of 659 potentially eligible articles were identified (Fig. 2). Thirty-one more articles were added by manual search or author's knowledge. Among them, a total of 652 articles were excluded. Thus, a total of 38 full-text articles were further assessed for eligibility. Of these, 25 more articles were excluded because they did not meet our inclusion criteria. Therefore, a total of 13 articles finally met our inclusion criteria for further study [7, 12, 18, 26–33, 35, 43].

Quality of studies

Overall, 10 studies (76.9%) were rated "high quality." Six articles were based on a prospective design, and seven more studies were retrospective single center (details in online resource 1).

Demographic data

A total of 316 adult patients with CMI without history of previous FMD were included in our systematic review. Of these, 127 were females (M:F ratio 1:1.41), and the mean age was 40.72 years (range 18–70). A total of 276 patients with CMI associated with syringomyelia were identified and evaluated. The mean follow-up was 33.91 months (range 6–78). Studies included in our systematic review are summarized in Table 2.

Clinical and radiological data

Spinal cord-related symptoms were the most common clinical presentation (254 patients). A total of 125 patients (49.2%, 95% CI 43.1–55.3%) complained of sensory disturbances, and 98 patients (38.6%, 95% CI 32.8–44.7%) presented with motor dysfunction (10 studies). Pain-related symptoms were reported in 139 patients. Tussive sub-occipital headache (Valsalva-related) and non-tussive

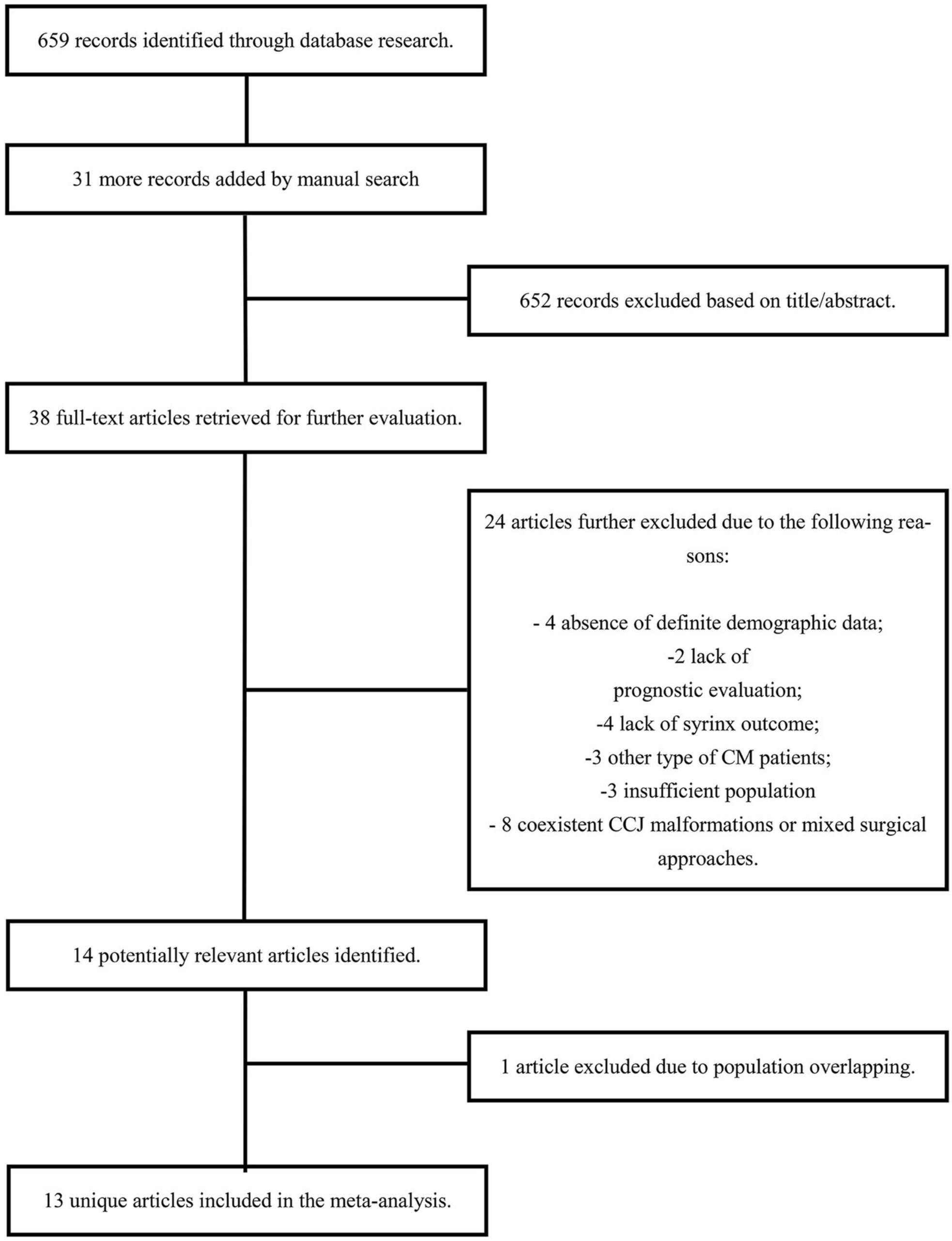

Fig. 2 PRISMA diagram detailing the specifics of the systematic literature review

headache were reported by 55 patients (39.6%, 95% CI 31.8–47.9%), and neck pain was described in 66 patients (47.5%, 95% CI 39.4–55.7%). Brainstem compression, cerebellar, and LCN-related symptoms were reported in 21, 36, and 14 patients respectively. Preoperative radiological data were available in four articles (122 patients). Syrinx location was cervical in 30 patients (24.6% 95% CI 17.8–33.0%), cervicothoracic in 73 patients (59.8% 95% CI 50.0–68.1%), thoracic in 4 patients (3.3%, 95% CI 1.0–8.4%), and holocord in 15 patients (12.3%, 95% CI 7.5–19.4%). Its mean extension was 8.8 vertebral levels (range 5–9). Detailed data are presented in Table 3.

Surgical approaches

FMDds was reported in 46 patients (14.6%, 95% CI 11.0–18.9%). FMDdo was reported in six studies. In total, 160 patients (50.6%, 95% CI 45.1–56.1%) underwent FMDdo. Finally, 110 patients (34.8%, 95% CI 30.0–40.2%) underwent FMDae.

Radiological outcome

Overall, the rate of post-operative radiological improvement at last follow-up was 81.1% (95% CI 73.3–88.9%; $p < 0.001$;

Table 3 Systematic review main data

	Raw data	Rate (95% CI)	*N* of articles
Demographic data			
N patients included in the analysis	316	–	13
N syrinx	276	87.3% (83.2–90.6%)	13
Sex			10
M	90/217	41.5% (35.1–48.1%)	
F	127/217	58.5% (51.9–64.9%)	
Age (mean, range)	40.72 (18–70)	–	11
Follow-up (mean, range)	33.91 (9–78)	–	8
Clinical presentation			
Headache	55/139	39.6% (31.8–47.9%)	
Neck pain	66/139	47.5% (39.4–55.7%)	
Sensory disturbances	125/254	49.2% (43.1–55.3%)	
Motor dysfunctions	98/254	38.6% (32.8–44.7%)	
Brainstem compression	21	–	5
Cerebellar	36	–	3
Lower cranial nerve	14	–	4
Preoperative symptom duration in months (mean, range)	31.36 (2–360)	–	6
Radiological features			
Syrinx location			4
Cervical	30/122	24.6% (17.8–33.0%)	
Cervico-thoracic	73/122	59.8% (50.0–68.1%)	
Thoracic	4–122	3.3% (1.0–8.4%)	
Holocord	15/122	12.3% (7.5–19.4%)	
Levels of syrinx extension (mean, range)	8.79 (5–9)	–	3

M male, *F* female

I^2 = 71.4%). Furthermore, a lower rate of post-operative radiological syrinx shrinkage was observed after FMDds (21/39 = 55.6% 95% CI 40.5–70.8%, I^2 = 0%), compared with FMDdo and FMDae (125/142 = 90%, 95% CI 85.1–94.8%, I^2 = 0% and 69/95 = 79.8%, 95% CI 61.7–98%, I^2 = 85.5%, respectively) ($p = 0.003$). Forest plots are shown in Fig. 3. On the other hand, the difference between extra-arachnoidal technique and arachnoid dissection was not significant in our analysis ($p = 0.307$).

No significant association arose between the rate of post-operative radiological outcome CSF-related complications, post-operative infections, tonsillar prolapse below C1, and the rate of cervical syringomyelia. Similarly, median follow-up did not affect the effect size, whereas longer preoperative duration of symptoms was associated with a lower rate of radiological improvement of syringomyelia (Table 4). Scatter plots are available in online resource 2.

Complications

Overall, CSF-related complications were reported in 19/297 patients (6.4% 95% CI 4.1–10%) (11 studies), and no significant relationship arose when considering surgical technique (FMDds, 1/45 = 2% 95% CI 0–12.6%; FMDdo, 14/146 = 9.6% 95% CI 5.7–15.6%; FMDae, 4/106 = 3.8% 95% CI 1.2–9.6%) ($p = 0.11$). The overall rate of post-operative infections was 10/270 = 3.7% (95% CI 1.9–6.8%) (10 studies) and was similar among sub-groups (FMDds, 2/44 = 4.5% 95% CI 0.4–16%; FMDdo, 6/118 = 5.1% 95% CI 2.12–10.9%; FMDae, 2/108 = 1.9% 95% CI 0.1–6.9%) ($p = 0.44$).

Study heterogeneity and publication bias

I^2 was greater than 50% for the rate of radiological outcome (71.4%). When considering surgical technique, heterogeneity was substantial for the FMDae sub-group (roughly 85%), and it was null for FMDds and FMDdo populations. No individual study significantly influenced outcomes, and the funnel plot followed by Egger's linear regression excluded the risk of publication bias ($p = 0.13$) (Fig. 4).

Discussion

FMD is widely accepted as the treatment of syringomyelia-CMI complex. Several details of the surgical procedure are

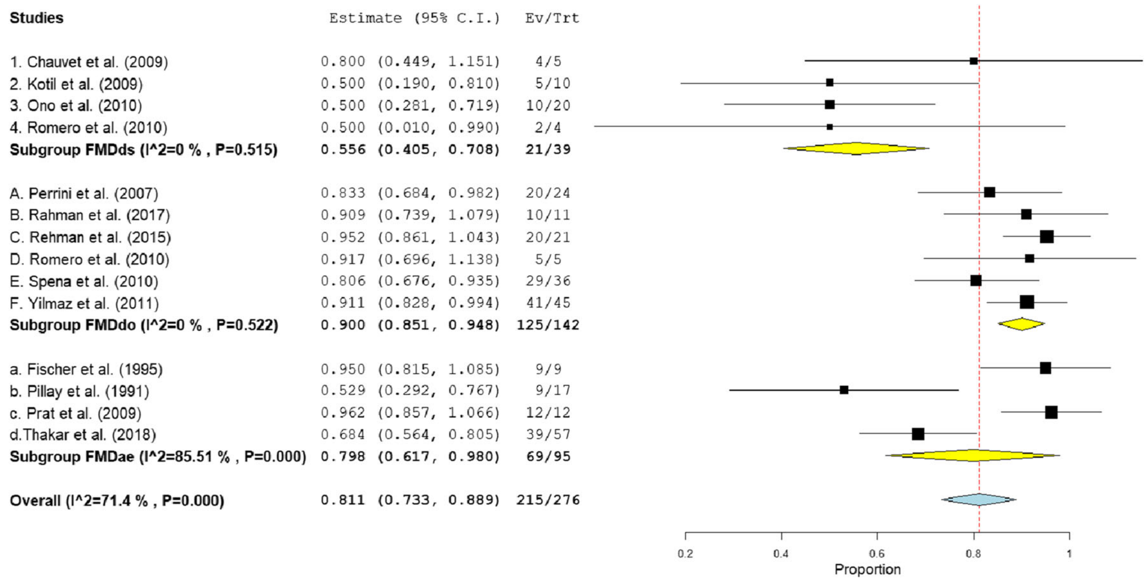

Fig. 3 Forest plot detailing sub-group analysis of pooled rate and 95% confidence intervals for the rate of syrinx radiological improvement after FMD. *Yellow diamond* sub-group pooled rate, *blue diamond* overall pooled rate. *ds* dural splitting, *do* dural opening, *ae* arachnoid exploration

under discussion and consensus on the ideal surgical treatment for this condition has not been reached. Pooling the results of 13 studies, our analysis provides representative data on the clinical and radiological outcome after FMD in patients with syringomyelia associated with CMI.

Post-operative changes of syringomyelia

The rate of post-operative reduction of syrinx size is quite variable in the literature ranging from 39 to 100% [25, 37]. The different surgical approaches and the heterogeneous populations reported in the clinical series may explain this high variability. In order to minimize the heterogeneity of our population, we did not select studies including patients with craniovertebral junction malformations and/or hydrocephalus. Our meta-analysis of 276 adult patients with CMI and syringomyelia found a post-operative reduction of syringomyelia in roughly 80% of cases with a mean radiological follow-up of 34 months (range 9–78). Among the analyzed confounders potentially affecting the rate of radiological shrinkage of syringomyelia, only the mean preoperative duration of symptoms was significant ($p = 0.03$). This result is in agreement with previous clinical studies and supports the concept that surgery should be offered early after diagnosis of syringomyelia [1, 5, 24]. One of the most relevant findings of this study is that patients undergoing FMD with splitting of the dura had significantly lower rates of syrinx reduction than patients treated with dural opening with or without arachnoid dissection (55.6% versus 79.8% and 90% [$p = 0.003$]). Dura splitting decompression was originally described by Isu et al. [16] in a small series of adult patients with syringomyelia associated with CMI. They reported a decrease in size of the syrinx in all patients and no complications. Subsequently, clinical series on decompression with incision of the outer dural layer in adults published rates ranging from 50 to 80% of syrinx reduction [7, 32]. In our analysis, a post-operative reduction of the syrinx after splitting of the dura was observed in 55.6% of patients (95% CI 40.5–70.8%) with low heterogeneity (I^2 = 0%). The lower number of post-operative syrinx shrinkage suggests that splitting of the dura may not be sufficient to completely relieve the occlusion of the free pulsatile CSF pathways across the craniovertebral junction [14]. Series recommending dural opening with or without arachnoid dissection reported higher rates of syrinx reductions ranging from 53 to 100% [28, 39]. Our study highlighted that rates of syrinx reduction were high among both groups of FMD with the extra-arachnoidal technique (90%, 95% CI 85.1–94.8%, $I^2 = 0\%$) and arachnoid dissection (79.8%, 95% CI 61.7–98%, $I^2 = 85.5\%$) without significant difference ($p = 0.307$). This finding neatly questions the issue of the

Table 4 Impact of potential confounders on the rate of radiological syrinx improvement ensuing FMD in random effects meta-regression

Variable	N. series	Intercept	Slope	*p* value
CSF-related complications	11	0.88	− 0.21	0.64
Post-operative infections	10	0.90	− 0.77	0.25
Median follow-up (months)	7	0.99	− 0.004	0.10
Tonsils descent below C1	5	0.99	− 0.006	0.15
Cervical syrinx	5	0.8	0.00	0.91
Mean duration of symptoms	5	4.05	− 0.008	0.03

FMD foramen magnum decompression

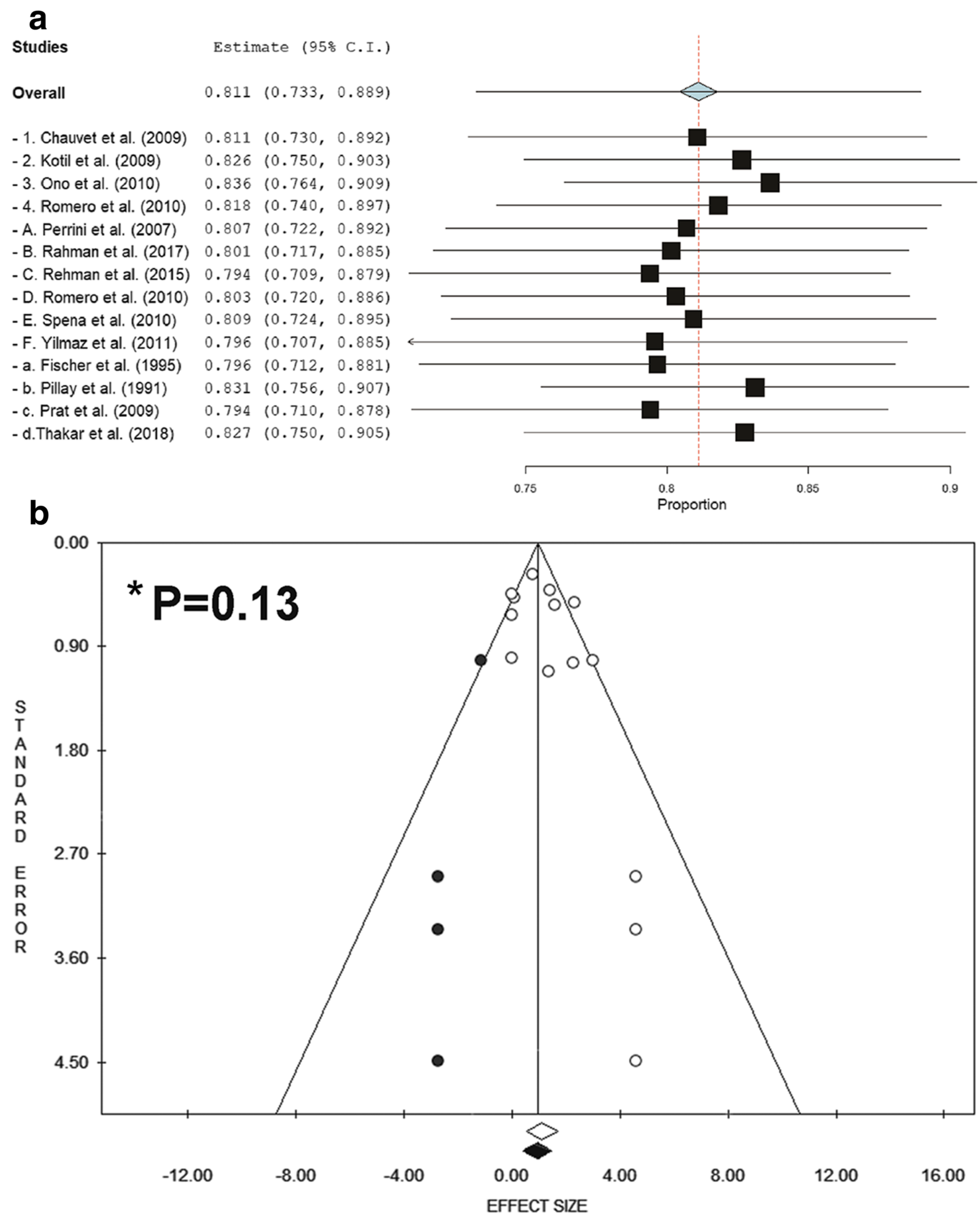

Fig. 4 **a** Leave-one-out sensitivity analysis for radiological outcome analysis ensuing FMD. **b** Funnel plot detailing publication bias among the included studies. *Asterisk* indicates statistical significance of Egger's linear regression test that excluded publication bias ($p = 0.13$)

pathophysiological role of intradural pathology in the occurrence of syringomyelia in the setting of CMI. Advocates for dissecting the arachnoid argue that intradural pathology, such as arachnoid veils or web and medialized tonsils, can partially or completely occlude the CSF flow channels through the foramen of Magendie, potentially leading to syringomyelia formation [9]. Tubbs et al. [36] found arachnoid veils in 9 (11.2%) out of 80 patients with CMI with an associated syrinx and recommended exploration of the fourth ventricle to ensure free CSF flow. However, these authors recognized that the intradural pathology cannot fully explain syringomyelia formation because arachnoid veils were not encountered on exploration of the fourth ventricle in most of the patients with syringes. In a recent clinical study, Dlouhy et al. [9] found obstructing arachnoid veils in 59.5% of patients with CMI associated with syringomyelia and suggested that this condition obstructs CSF flow out of the fourth ventricle with resultant downward herniation and piston-like effect of the tonsils. Similarly, Klekamp [17] in a clinical study of 371 FMDs found that the grade of arachnoid pathology correlated with the rate of associated syringomyelia and recommended arachnoid opening in every decompression with the aim to restore the CSF pathways. However, Wetjen et al. [39], in a prospective study on 29 consecutive patients with syringomyelia-CMI complex operated on with the extra-arachnoidal technique, found that syringomyelia decreased in diameter and length during the radiological follow-up in all patients suggesting that the underlying pathophysiology can be reversed without addressing the intradural pathology. In fact, in a seminal anatomical study on the foramen of Magendie, Barr [4] found that cases of imperforate foramina of Luschka and cases of shelf-like midline membranous projections blocking the median aperture are common in otherwise healthy humans. According to these findings, the pathophysiological role of intradural pathology in syringomyelia formation remains elusive and the role of intradural exploration in the treatment of syringomyelia-CMI complex continues to be under discussion.

Pathogenesis of syringomyelia associated with Chiari type I malformation and surgical implications

The majority of the hypotheses dealing with the pathogenesis of syringomyelia associated with CMI supports the existence of an altered cranio-spinal CSF dynamics. With the "hydrodynamic theory" of syringomyelia, Gardner postulated that incomplete opening of the outlets of the fourth ventricle leads the CSF being forced into the central canal of the spinal cord with resultant syringomyelia formation [13]. Accordingly, in order to treat the intradural pathology, he proposed plugging the obex as the most physiological surgical option for syringomyelia in patients with CMI. Subsequently, Williams proposed that the existence of cranio-spinal CSF pressure dissociation in patients with CMI promotes the CSF flow from the fourth ventricle to the central canal [40]. Interestingly, Williams suggested to avoid plugging of the obex although he maintained a general agreement upon the necessity of intradural manipulation including arachnoid dissection and amputation of cerebellar tonsils [41]. Subsequently, many clinical and experimental studies suggested that the syrinx fluid could originate from the subarachnoid CSF entering into the spinal cord through the perivascular or interstitial spaces [3, 22, 25]. Oldfield et al. proposed that the mechanism for the development of syringomyelia lies in a piston-like effect of the herniated cerebellar tonsils on the partially enclosed spinal CSF with resultant increased subarachnoid pulse pressure [25]. According to this pathophysiological hypothesis, an extra-arachnoidal cranio-cervical decompression can effectively reverse the mechanism of syringomyelia formation and progression dismissing the role of intradural pathology. The concept that syringomyelia arises from accumulation of extracellular fluid coming from the spinal cord microcirculation originally suggested by Tannennerg in 1924 [34] and Liber and Lisa in 1937 [20] has been recently reproposed. Koyanagi and Houkin [19] hypothesized that in patients with CMI and syringomyelia, the reduced compliance of the posterior spinal veins due to the decreased compliance of the spinal subarachnoid space interferes with the absorption mechanism of the extracellular fluids through the intramedullary venous channels. The resultant enlargement of the central canal and interstitial edema promotes syringomyelia formation. Similarly to other hypotheses, also in the latter, the obstruction of the foramen magnum remains the initiating factor promoting syringomyelia formation and progression, whereas the role of intradural pathology remains poorly investigated.

Treatment-related complications

One of the main concerns of surgical treatment of syringomyelia associated with CMI is the risk of CSF-related complications, including CSF leakage, pseudomeningocele, aseptic meningitis, and hydrocephalus. Scanty information is available in the literature on surgical morbidity after FMD for CMI. In a recent retrospective study, Klekamp [17] reported a complication rate of 21.8% and the most common complication was CSF fistula. Previous meta-analyses reported a higher likelihood of CSF-related complications after FMD with duraplasty compared with FMD without duraplasty [6, 21]. Lin et al. [21] found a significant lower relative risk of overall complications in the group of FMD without duraplasty (RR, 0.78, 95% CI, 0.66–0.93; $p < 0.05$) compared with FMD with duraplasty, although no significant difference was found in the rate of wound infection between the two groups. The main limitation of these meta-analyses is the heterogeneity of the included population. In fact, duraplasty is usually performed in both FMD with extra-arachnoidal technique and arachnoid dissection and series exist in which extra-arachnoidal technique is accomplished without duraplasty. In our meta-analysis, which identified three different subgroups of FMD, CSF-related complications occurred in 6.4% (95% CI 4.1–10%) of patients without significant differences among the surgical techniques. The occurrence of CSF leakage after FMD with incision of the external layer of the dura is the result of accidental opening of both the inner dural layer and the arachnoid [18]. Similarly, CSF-related complications after extra-arachnoidal technique are related to inadvertent arachnoidal lacerations and tears, as well as incompetent duraplasty [27].

Strength and limitations

Our study has several limitations. Series are often retrospective studies and small single-institution experiences. Because of the small number of cases, the comparison between subgroups may not provide a comprehensive representation of outcomes difference among different surgical approaches. In addition, the series included in our study range was over a wide temporal period, and the surgical management of CMI changed in this time period. Furthermore, imaging technology has become more sensitive in detecting post-operative changes of syringomyelia. Due to the relatively short follow-up, our analysis is unable to comment on the long-term results of different techniques of FMD. Particularly, it is important to point out that it was impossible to assess the rate of repeat surgery for syringomyelia recurrence, due to insufficient data. In addition, we were not able to analyze the effect of arachnoid pathology on the radiological outcome due to insufficient data. Furthermore, three confounders (tonsillar prolapse below C1, rate of cervical syringomyelia, and median duration of symptoms) were available only in five studies, increasing the risk of selection bias. However, although retrospective data are low in quality, our meta-analysis is the best available evidence to assess the efficacy of different FMD techniques in syringomyelia resolution of patients with CMI.

Conclusions

Our meta-analysis found a post-operative reduction of syringomyelia in roughly 80% of adult patients after FMD for CMI. That rates of syrinx reduction did not differ significantly among the groups of decompression with the extra-arachnoidal technique and arachnoid dissection, whereas patients undergoing decompression with splitting of the dura had significantly lower rates of syrinx reduction. A large prospective, randomized study is required to conclusively answer the question on how the operation should be correctly tailored in adult patients with syringomyelia associated with CMI.

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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 consent is not required.

Informed consent The nature of this article did not require informed consent.

Research involving human participants and/or animals This article does not contain any studies with human participants or animals performed by any of the authors.

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