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Clinical and Radiological Outcomes of Dura-Splitting versus Duraplasty Techniques in Pediatric Chiari I Malformation: A Systematic Review and Meta-Analysis

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Abstract

Type I Chiari malformation is a developmental anomaly with various proposed surgical techniques for its management. The dura-splitting technique is a less invasive approach and involves the resection of the outer layer of the dura while sparing the internal layer. While this less-known approach may minimize the complication rates, there are concerns about its efficacy and outcome. Therefore, we have performed a systematic review and meta-analysis of available data on clinical and radiological outcomes of this technique in the pediatric population and compared them to the foramen magnum decompression and duraplasty technique.

We have followed the Meta-analysis Of Observational Studies in Epidemiology guidelines in this review. Based on our predefined search strategy, we performed a systematic database search. Subsequently, the article screening process was done based on defined inclusion/exclusion criteria. Following the quality assessment of included studies, two authors performed data extraction. Finally, the extracted data were summarized and presented in form of tables. Forest plots were used to demonstrate the results of the meta-analysis.

A review of 8 included studies consisting of 615 patients revealed the significant advantage of the dura-splitting technique in terms of shorter operation duration and hospital stay. The recurrence rate and clinical and radiological outcomes were almost similar between the two surgical techniques. Complication rates were significantly lower in the dura-splitting technique.

Dura-splitting can be an effective and safe approach for the management of pediatric Chiari I malformation. However, these results are mostly extracted from observational studies and future randomized controlled trials are recommended.

Keywords
- Chiari malformation type I
- dura-splitting
- duraplasty
- outcome
- pediatric

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Introduction

Chiari malformation type I (CM-I) is a condition in which herniation of the cerebellar tonsils more than 5mm below the level of the foramen magnum occurs with the presence of concomitant developmental anomalies of the posterior fossa.[1] The resultant obstruction of cerebrospinal fluid (CSF) flow at the craniocervical junction may cause syringomyelia formation.[2] Although the clinical presentation of this condition is usually in the third and fourth decades of age, CM-I is also commonly diagnosed in the pediatric population. An array of techniques has been introduced for the surgical management of this malformation. The technique that the majority of neurosurgeons are familiar with is suboccipital craniectomy followed by duraplasty. Recently, less invasive surgical techniques without duraplasty such as posterior fossa decompression alone or with the splitting of the outer layer of dura have shown promising outcomes and low complication rates. In the duraplasty technique, dura opening and expansion using autograft or allograft is performed following the suboccipital craniectomy. In some instances, manipulation of intradural structures such as tonsillar resection or arachnoid adhesiolysis may also be performed in this technique. In contrast, the less invasive dura-splitting technique includes suboccipital craniectomy followed by resection of the outer layer of the dura without disturbing the inner layer. There are a few studies in the literature about the clinical and radiological outcome of the dura-splitting technique compared to other common surgical techniques in the management of pediatric CM-I malformation. But the strengths and weaknesses of this surgical technique are not elucidated yet. We recently performed a systematic review and meta-analysis on this topic in the adult population, which interestingly demonstrated acceptable and favorable outcomes of the dura-splitting technique in adults.[3] So, we decided to perform a well-conducted systematic review specifically dedicated to the pediatric population to also address this controversy in the management of pediatric CM-I.

Materials and Methods

Literature Search and Screening

We have conducted this review based on the review protocol registered in the PROSPERO (International Prospective Register of Systematic Reviews) database with the registration number CRD42019134771.[4] Because most of the included studies in this review are observational studies, we implemented Meta-analysis Of Observational Studies in Epidemiology (MOOSE) reporting guidelines for systematic reviews and meta-analyses of observational studies in this review.[5] Initially, we designed a comprehensive search strategy based on our defined PICO (Population, Intervention, Comparator, Outcome) including different spellings of "Chiari malformation type I" AND "Dura-splitting" as keywords (►**Supplementary Material 1**). The literature search was performed using Scopus, Medline, and Cochrane databases. Our predefined inclusion criteria included: Trials and observational studies with a study population consisting of less than 18 years old patients with a definite diagnosis of CM-I, AND divided into at least two dura-splitting and duraplasty groups AND with available details on clinical or radiological outcome measures, intraoperative and postoperative parameters or complication rates. Our exclusion criteria included the following: (a) Studies included patients with recurrent Chiari. (b) Studies that also included patients with other concomitant craniospinal anomalies. (c) Numerous studies published by one author on a single population (all articles except the last article were excluded). The bibliographic data of included articles were also evaluated to find other related articles.

After the completion of the article search process, the titles and abstracts of the articles were screened in terms of compliance with the inclusion and exclusion criteria by two authors (EK, AG), independently and without the knowledge of the authors of the articles and the journal in which the articles were published. Any conflicts between the two authors in the first stage were resolved by discussion and in case of no agreement a third opinion was sought from the senior author (AT). Then the full text of the approved articles was prepared and subjected to a similar review process.

Quality Assessment

Two researchers (AT, HR) independently used the critical appraisal tools provided by the National Heart, Lung, and Blood Institute (NHLBI), which is an National Institute of Health (NIH)-related institute, to assess the quality of the included studies. Any discrepancies between the scorings of the two authors were put into discussion.

Data Extraction

To reduce errors in the data extraction process, this critical process was accomplished by two independent authors (AG, AS) using a predefined data extraction tool (►**Supplementary Material 2**). Our data of interest to extract were categorized as characteristics of the included studies such as study design, groups, and sample sizes, demographic data, clinical or radiological outcome measures, intraoperative parameters such as blood loss and duration of surgery, and also complication rates. The extracted data by the two authors were compared to each author for any discrepancy and the differences were rechecked to ensure a final error-free datasheet to work with.

Statistical Analysis

We used the Comprehensive Meta-Analysis (CMA) software to perform the meta-analysis of extracted data. We have used risk ratio (RR) and standardized mean difference (SMD) as effect sizes for meta-analysis of categorical and scale variables, respectively. Higgins index (I^2) was used for the assessment of heterogeneity of data.[6] The threshold for significant heterogeneity was considered as I^2 of more than 50% and the statistical significance was defined as p-value less than 0.05. We used the fixed and random effects model for meta-analysis of studies without and with significant heterogeneity, respectively.[7] The results of the meta-analysis were graphically demonstrated in form of forest plots.

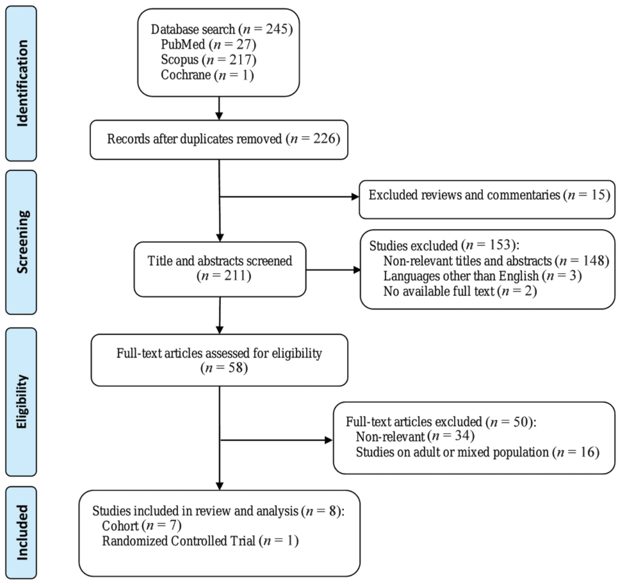

Fig. 1 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) chart describing the flow of article screening procedure.

Results

Included Studies

Our initial search resulted in finding 226 articles. After excluding review articles and commentaries, the screening of titles and abstracts revealed 58 relevant articles. Among the 58 carefully screened full-text articles, eight studies were finally approved for inclusion in our review. These studies included seven cohort studies[8–14] and one randomized controlled trial[15] (►**Fig. 1**). An overview of the included studies is presented in ►**Table 1**.

Quality Assessment

Based on the quality assessment performed by the two authors, seven studies had a fair quality,[8,9,11–15] while only one study showed an overall quality of less than moderate (►**Fig. 2**).[10] The absence of any sample size justification and not presenting any information about whether or not the outcome assessors were blinded were among the most common drawbacks of the included studies (►**Supplementary Material 3** and **4).**

Demographic Data

Our review cohort included a total of 615 pediatric CM-I patients. Of these, 313 patients had been surgically treated with the dura-splitting technique, while the foramen magnum decompression and duraplasty procedure had been performed on the other 302 patients. Patients' age ranged between 0.5 and 18 years (mean: 9.7 years) and the mean age was lower in the dura-splitting compared to the duraplasty group (9.3 vs. 11.5). Overall M:F ratio was 1:1.1 and this ratio was almost similar in both groups (►**Table 2**).

Clinical Findings

The most common presenting symptom was a pain in 56.8% of the total review cohort in the forms of suboccipital, cervical, or extremity pain. Other reported signs/symptoms were sensory deficit, motor deficit, cranial nerve dysfunction, visual symptoms, and cerebellar signs in order of decreasing prevalence (►**Table 3**).

Preoperative Imaging Findings

The prevalence of preoperative syrinx was 52% in the total review cohort. This prevalence was higher among patients who had been treated with the duraplasty technique (81.6% compared to 29.3% in the dura-splitting group). The size of preoperative syrinx was reported in a few studies with the average of extension throughout 9.2 spinal levels. Mean tonsillar descent below the foramen magnum was calculated as 11.1 mm in the dura-splitting group with no considerable difference compared to the duraplasty group. The mean prevalence of scoliosis was 29.5% and almost 10.5% of patients had been diagnosed with concomitant hydrocephalus (►**Table 4**).

Operative Findings

The mean blood loss volume during surgery was almost similar in both groups, while the dura-splitting technique benefited from a considerably less operation duration and hospital stay compared to duraplasty (►**Table 5**).

Clinical and Radiological Outcome

The mean follow-up duration was 14.5 months (1–74 m) in the dura-splitting group, while it was relatively higher in the duraplasty cohort (18.3 months). The average rate of postoperative clinical improvement was calculated as 64.7% among the patients in the dura-splitting group that was not significantly different from the 63.5% clinical improvement rate in the duraplasty cohort. In a few studies that reported the rates of clinical stability and worsening, the clinical stability rate seems to be higher in the dura-splitting cohort, while the clinical worsening rate is reported to be higher amongst the duraplasty group patients. The pain and motor deficits were the most responsive signs/symptoms to surgery, but the cerebellar and sensory signs were less amenable to recovery following both surgical techniques. The pooled estimates of recurrence/reoperation rates reported by four studies were 9.6 and 7.1% for the dura-splitting and duraplasty cohorts, respectively (►**Table 6**). The postoperative rate of radiological improvement (the reduction of syrinx size or its complete resolution) was considerably high in both groups with almost similar rates (76.8 vs. 75.8%). Postoperative improvement of the tonsillar descent was relatively higher in the duraplasty (85.4%) compared to the dura-splitting cohort (75%). A summary of all postoperative radiological data is presented in ►**Table 7**.

Complications

The duraplasty technique was associated with a considerably higher overall complication rate (27.4%) compared to the dura-splitting technique (4.9%). The occurrence of complications related to CSF (i.e., leakage, pseudomeningocele, aseptic meningitis) was 20% in the duraplasty group as opposed to a very low rate of 0.7% in the dura-splitting

Table 1 Summary of included studies

Author, year	Study type	Period	N	Study groups (n)	Conclusions	Limitations
Limonadi and Selden, 2004[8]	Cohort	2000–2002	24	Dura-splitting (12) Duraplasty (12)	Safety and efficacy of dura-splitting in patients without syringomyelia	Small sample size, short follow-up, lack of clinical follow-up data
Navarro et al, 2004[9]	Cohort	1981–2001	109	Dura-splitting (56) Duraplasty (53)	Dura-splitting is recommended as the initial surgical procedure in patients with or without syringomyelia with fewer complications and comparable success rates	The retrospective setting, lack of intraoperative data
Galarza et al, 2007[10]	Cohort	1997–2002	60	Dura-splitting (20) Duraplasty (40)	Duraplasty was equally effective compared to dura-splitting. Among patients with syringomyelia, the tonsillar reduction was associated with a significantly better outcome	The retrospective setting, short follow-up, lack of intraoperative data
Litvack et al, 2013[11]	Cohort	2000–2009	110	Dura-splitting (63) Duraplasty (47)	Dura-splitting was equally effective, safer, and with lower cost for treatment of patients without syringomyelia	The retrospective setting, short follow-up, lack of radiological follow-up data
Pomeraniec et al, 2016[12]	Cohort	2004–2013	25	Dura-splitting (14) Duraplasty (11)	The conservative management of CM-I patients does not result in significant clinical or radiological progression	The retrospective setting, small sample size, lack of intraoperative data
Pisapia et al, 2017[14]	Cohort	2004–2014	189	Dura-splitting (98) Duraplasty (91)	Equal clinical outcomes and recurrence rates in both techniques support the dura-splitting approach as the first-line surgical option	The retrospective setting, short follow-up, lack of intraoperative data
Grahovac et al, 2018[13]	Cohort	2007–2014	16	Dura-splitting (10) Duraplasty (6)	Duraplasty tends to have a lower recurrence rate and better long-term outcome compared to dura-splitting despite higher complication rates	The retrospective setting, small sample size, lack of intraoperative data, lack of radiological follow-up data
Jiang et al, 2018[15]	RCT	2011–2015	82	Dura-splitting (40) Duraplasty (42)	Dura-splitting produces comparable radiologic and clinical outcomes with a lower risk of complications compared to duraplasty	Lack of clinical follow-up data
Sum	Cohort (7) RCT (1)	1981–2015	615	Dura-splitting (313) Duraplasty (302)	—	—

Abbreviations: CM-I, Chiari malformation 1; RCT, randomized controlled trial.

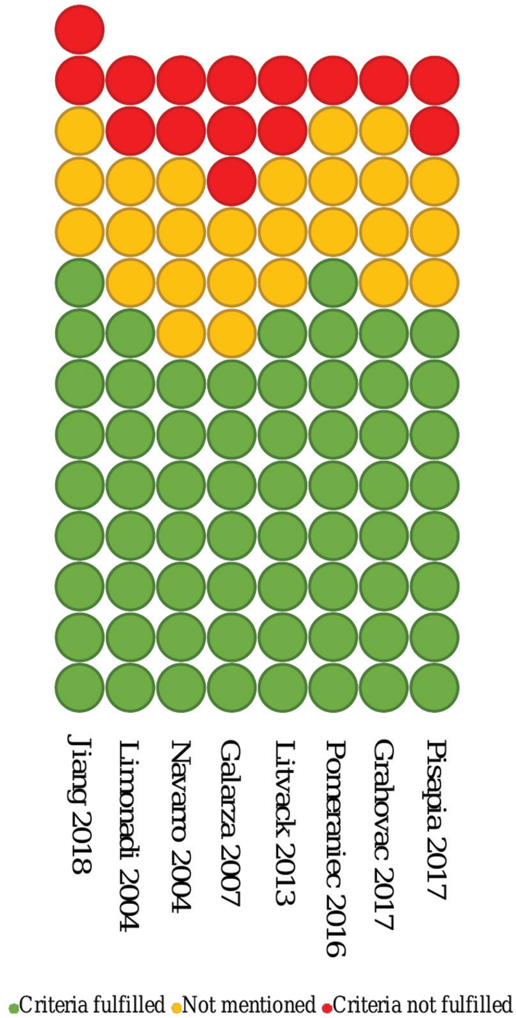

Fig. 2 Graphical presentation of quality assessment of included articles.

group. Moreover, the duraplasty technique suffered from a considerably higher infection rate (4.7%) compared to dura-splitting (2.8%). The occurrence rate of the new postoperative neurological deficit was 2.8% in the duraplasty group while there were no reported new deficits following the dura-splitting technique (►**Table 8**).

Meta-Analysis

Operative Parameters

There was a statistically significant difference between hospital stay duration of patients in the two review groups with patients of the dura-splitting cohort having significantly shorter hospital stay duration (SMD: −0.56; 95% confidence interval [CI]: −0.90 to 0.22; $p=0.001$; $I^2=58.7$; ►**Fig. 3**). A comparison of the mean duration of surgery between two surgical techniques demonstrated significantly lower operation duration in the dura-splitting group (SMD: −1.25; 95% CI: −2.17 to 0.34; $p=0.007$; $I^2=87.7$; ►**Fig. 3**). Interestingly, there was no significant difference between the mean intraoperative blood loss volume of the two surgical techniques (SMD: −0.01; 95% CI: −0.39 to 0.36; $p>0.05$; $I^2=0$; ►**Fig. 3**).

Clinical Follow-Up

The clinical improvement rate did not show a statistically significant difference between the two surgical techniques (RR: 0.99; 95% CI: 0.76–1.30; $p>0.05$; $I^2=55.9$; ►**Fig. 4**). But, the mean rate of clinical stability was significantly higher in the dura-splitting group (RR: 2.96; 95% CI: 1.37–6.41; $p=0.006$; $I^2=0$; ►**Fig. 4**). Worsening of symptoms was reported only in one included study and there was no significant difference between the two surgical techniques in this regard (RR: 0.39; 95% CI: 0.08–1.76; $p>0.05$; ►**Fig. 4**). On the other hand, the meta-analysis had failed to show significantly different rates of recurrence/reoperation between dura-splitting and duraplasty techniques (RR: 1.33; 95% CI: 0.72–2.45; $p>0.05$; $I^2=0$; ►**Fig. 4**).

Radiological Follow-Up

A meta-analysis of available data from four included studies that have reported postoperative syrinx resolution/improvement rate could not reveal any significant difference between

Table 2 Summary of demographic data

Author, year	Age (y), mean (range)			Male (%)			Female (%)		
	Overall	DS	DP	Overall	DS	DP	Overall	DS	DP
Limonadi and Selden, 2004[8]	9.2 (2–18)	7.6 (2–14)	10.8 (3–18)	46	42	50	54	58	50
Navarro et al, 2004[9]	8.2 (0.5–18)	–	–	–	–	–	–	–	–
Galarza et al, 2007[10]	10 (1–18)	–	–	50	–	–	50	–	–
Litvack et al, 2013[11]	9.1 (3–15)	8.3 (3–14)	10.4 (6–15)	54	54	53.9	46	46	46.1
Pomeraniec et al, 2016[12]	10.2 (1.8–18)	10.9 (2.5–18)	9.8 (1.8–16)	56	57.1	54.5	44	42.8	45.4
Pisapia et al, 2017[14]	10 (6–16)	9 (6–16)	12 (6–16)	42	42	42	58	58	58
Grahovac et al, 2018[13]	1.7 (0.5–2.9)	1.6 (0.5–2.9)	1.7 (0.9–2.5)	50	50	50	50	50	50
Jiang et al, 2018[15]	13.8 (10–18)	13.6 (10–18)	13.9 (10–18)	50	50	50	50	50	50
Pooled estimates	9.7 (0.5–18)	9.3 (0.5–18)	11.5 (0.9–18)	47.8	47.6	47.3	52.1	52.3	52.6

Abbreviations: DP, duraplasty; DS, dura-splitting.

Table 3 Summary of presenting symptoms

Author, year	Pain[a] (%)	Cranial nerve involvement[b] (%)	Visual symptoms (%)	Ataxia (%)	Sensory symptoms[c] (%)	Vertigo (%)	Sensory deficit (%)	Motor deficit (%)
Limonadi and Selden, 2004[8]	71	21	8	29	29	—	—	33
Navarro et al, 2004[9]	48.6	15.6	—	—	—	—	—	—
Galarza et al, 2007[10]	75	5	—	13.3	8.3	5	8.3	20.8
Litvack et al, 2013[11]	74	15	—	—	—	—	—	28
Pomeraniecet al, 2016[12]	76	32	16	8	20	8	20	12
Grahovac et al, 2018[13]	75	62.5	—	6.2	—	—	—	6.2
Jiang et al, 2018[15]	18.3	—	—	2.4	1.2	1.2	34.1	23.2
Pooled estimates	56.8	17.4	12.2	9.6	9.4	3.5	22.7	17.6

[a]Including all suboccipital, cervical, and extremity pains.
[b]Including Gag weakness, dysphagia, and hoarseness.
[c]Including numbness, paresthesia, and hypesthesia.

the two surgical techniques (RR: 0.95; 95% CI: 0.81–1.10; $p > 0.05$; $I^2 = 18.2$; ►**Fig. 5**). Similarly, the mean rate of postoperative syrinx size stability did not differ significantly between the two groups (RR: 1.73; 95% CI: 0.76–3.94; $p > 0.05$; $I^2 = 0$; ►**Fig. 5**). The two surgical techniques have also shown an almost similar syrinx progression rate in the included studies (RR: 0.84; 95% CI: 0.30–2.32; $p > 0.05$; $I^2 = 0$; ►**Fig. 5**).

Complications

The overall complication rate of the dura-splitting technique was significantly lower compared to the duraplasty technique (RR: 0.20; 95% CI: 0.10–0.38; $p < 0.001$; $I^2 = 0$; ►**Fig. 6**). Among all complications, the mean rate of CSF-related events was considerably higher in the duraplasty technique (RR: 0.07; 95% CI: 0.02–0.23; $p < 0.001$; $I^2 = 0$), while the infection rate was almost similar between the two surgical techniques (RR: 0.59; 95% CI: 0.23–1.47; $p > 0.05$; $I^2 = 0$; ►**Fig. 6**). Patients who had been operated on using either dura-splitting or duraplasty techniques had shown low rates of postoperative neurological deficits with no significant difference between the two techniques in this regard (RR: 0.29; 95% CI: 0.03–2.67; $p > 0.05$; $I^2 = 0$; ►**Fig. 6**).

Discussion

Foramen magnum decompression constitutes the mainstay of management in CM-I. However, there is an array of surgical techniques available to achieve this purpose, and neurosurgeons should be aware of the pros and cons of each technique and weigh them during the surgical management of CM-I patients. Nevertheless, the available literature in this regard has failed to reach a consensus on the superiority of a specific technique in terms of clinical or radiological outcomes over others till now. There are multiple reports on the promising results of dura-splitting as the latest and less-known surgical technique introduced for the safe and efficient surgical management of CM-I patients. However, we tried to present a higher level of evidence by systematically reviewing and pooling data extracted from eight available studies.

Preoperative Parameters

The minimal confounding effect of the preoperative variables on the outcome measures can be ascertained by the fact that the two review cohorts (dura-splitting and duraplasty) were considerably similar in terms of preoperative parameters such as demographic factors, nature and duration of presenting symptoms, and preoperative imaging findings. However, the slightly lower age of patients in the dura-splitting cohort may reflect the preference of neurosurgeons toward pursuing a more aggressive approach such as duraplasty in older patients. This orientation originates from the biomechanical concept of the loss of elasticity of the inner dural layer with increasing age. Contradictory to this concept, we have recently reported almost similar clinical and radiological patient outcomes following the dura-splitting technique compared to duraplasty even in the adult population.[3] Another expected discrepancy between the two groups of the review was a considerably higher proportion of patients presenting with a preoperative syrinx in the duraplasty cohort. As a nonwritten rule, neurosurgeons tend to go with the duraplasty technique for the management of CM-I patients with coexisting syringomyelia. A justification behind this tendency is that multiple studies are demonstrating the superiority of the dura-opening techniques over foramen magnum bony decompression (without dura-splitting) in terms of postoperative syrinx resolution.[16–22] In the dura-splitting technique, the expansion of the inner dural layer may result in the restoration of normal CSF flow at the craniocervical junction more than it can be achieved by the bony decompression alone, given the fact that the outer dural layer has less elastic and expansile characteristics and limits the achievable dural expansion. Therefore, it is theoretically

Table 4 Summary of preoperative radiological parameters (numbers in parentheses are presenting the reported ranges)

Author, year	Syrinx (%)			Scoliosis (%)			Hydrocephalus (%)			Tonsillar descent (mm) mean (range)			Syrinx length (levels) mean (range)		
	Overall	DS	DP	Overall	DS	DP	Overall	DS	DP	Overall	DS	DP	Overall	DS	DP
Limonadi and Selden, 2004[8]	50	0	100	4	0	8	–	–	–	–	–	–	–	–	–
Navarro et al, 2004[9]	44	16	73.5	12.8	–	–	–	–	–	–	–	–	–	–	–
Galarza et al, 2007[10]	40	–	–	13.3	–	–	10	–	–	–	–	–	–	–	–
Litvack et al, 2013[11]	44.5	7.9	93.6	–	–	–	–	–	–	11.4 (5–28)	9.7 (5–16)	12.5 (5–28)	–	–	–
Pomeraniec et al, 2016[12]	64	64.2	63.6	–	–	–	–	–	–	19 (8–37)	20 (8–31)	17.7 (8–37)	6.7 (2–14)	7 (2–14)	6.4 (2–12)
Pisapia et al, 2017[14]	47	22	73	–	–	–	–	–	–	12 (8–16)	12 (8–17)	12 (8–16)	–	–	–
Grahovac et al, 2018[13]	6.2	10	0	0	0	0	12.5	0	33.3	9.2 (4–16)	8.2 (4–12)	10.9 (7–16)	–	–	–
Jiang et al, 2018[15]	100	100	100	76.8	82.5	71.4	–	–	–	9.4 (5–22)	9 (5–17)	9.9 (5–22)	10 (1–19)	9.3 (1–18)	10.7 (3–19)
Pooled estimates	52	29.3	81.6	29.5	53.2	51.6	10.5	0	33.3	11.6 (4–37)	11.1 (4–31)	11.9 (5–37)	9.2 (1–19)	8.7 (1–18)	9.8 (2–19)

Abbreviations: DP, duraplasty; DS, dura-splitting.

Table 5 Summary of operative parameters (numbers in parentheses are presenting the reported ranges)

Author, year	Blood loss (cc) mean (range)			Operation duration (min) mean (range)			Hospital stay (d) mean (range)		
	Overall	DS	DP	Overall	DS	DP	Overall	DS	DP
Limonadi and Selden, 2004[9]	76.5 (25–300)	73 (25–300)	80 (25–250)	134	99	169	3.3	3	3.75
Litvack et al, 2013[11]	–	–	–	132.5 (36–244)	105.5 (36–175)	168.9 (94–244)	2.5 (1–5)	2.4 (1–4)	2.8 (1–5)
Pomeraniec et al, 2016[12]	–	–	–	–	–	–	3.4 (2–5)	3.1 (2–5)	3.8 (2–5)
Pisapia et al, 2017[14]	–	–	–	–	–	–	3 (1–63)	2 (1–63)	4 (2–61)
Jiang et al, 2018[15]	70 (15–400)	70 (15–200)	69 (15–400)	142 (45–510)	119 (45–220)	166 (51–510)	8.9 (6–21)	8.1 (6–11)	9.8 (7–21)
Pooled estimates	71.4 (15–400)	70.6 (15–300)	71.4 (15–400)	136.2 (36–510)	109.5 (36–220)	167.7 (51–550)	4 (1–63)	3.3 (1–63)	4.8 (1–61)

Abbreviations: DP, duraplasty; DS, dura-splitting.

Table 6 Summary of clinical outcomes (numbers in parentheses are presenting the reported ranges)

Author, year	FU duration (month)			Clinical status									Recurrence Reoperation (%)			Signs or symptoms improvement (%)					
	mean (range)			Improved (%)			Stable (%)			Worsened (%)						Pain		Motor		Cranial nerve	
	Overall	DS	DP	All	DS	DP	All	DS	DP	All	DS	DP	All	DS	DP	DS	DP	DS	DP	DS	DP
Limonadi and Selden, 2004[8]	15.3 (3–30)	15.7 (4–27)	14.8 (3–30)	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–
Navarro et al, 2004[9]	27.6 (2–117)	–	–	70.6	72.2	68.4	23.8	–	–	3.6	–	–	11.9	14.2	9.4	–	–	–	–	–	–
Galarza et al, 2007[10]	21 (12–120)	–	–	70	47.3	82.5	30	52.5	17.5	–	–	–	–	–	–	–	–	–	–	–	–
Litvack et al, 2013[11]	16 (1–36)	13.9 (1–32)	18.7 (2–36)	–	–	–	–	–	–	–	–	–	0.9	1.6	0	90	91.4	92.9	88.2	100	100
Pomeraniec et al, 2016[12]	60.8 (29–92)	45.8 (17–74)	80 (55–105)	72	78.5	63.6	4	7.1	0	24	14.2	36.3	–	–	–	–	–	–	–	–	–
Pisapia et al, 2017[14]	2 (1–75)	2 (1–68)	3 (1–75)	53.4	57.9	48.8	–	–	–	–	–	–	8.4	8.1	7.6	92	79	–	–	72	81
Grahovac et al, 2018[13]	67.2 (36–120)	–	–	100	100	100	–	–	–	–	–	–	43.7	50	33.3	100	100	–	–	–	–
Jiang et al, 2018[15]	35.6 (24–56)	35.2 (24–56)	36 (24–48)	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–	–
Pooled estimates	19.9 (1–120)	14.5 (1–74)	18.3 (1–105)	63.9	64.7	63.5	23.1	33.3	13.7	7.4	14.2	36.3	8.7	9.6	7.1	88.8	92.3	92.9	88.2	73.6	83

Abbreviations: DP, duraplasty; DS, dura-splitting; FU, follow-up.

Table 7 Summary of radiological outcomes

Author, year	Radiological improvement (%)	Postoperative syrinx size changes									Tonsillar descent improvement (%)		
		Resolution/ Improvement (%)			Stable (%)			Progression (%)					
		All	DS	DP	All	DS	DP	All	DS	DP	All	DS	DP
Limonadi and Selden, 2004[8]	–	–	–	100	–	–	0	–	–	0	–	–	–
Navarro et al, 2004[9]	66	65.7	–	–	28.9	–	–	–	–	–	–	–	–
Galarza et al, 2007[10]	–	64.2	40	77.7	35.7	60	22.2	–	–	–	–	–	–
Pomeraniec et al, 2016[12]	87.5	87.5	100	71.4	–	–	–	31.2	33.3	28.5	85.7	92.8	71.4
Pisapia et al, 2017[14]	–	59.7	60	59.6	20.8	26.6	19.2	19.4	13.3	21.1	–	–	–
Grahovac et al, 2018[13]	–	–	–	–	–	–	–	–	–	–	50	30	83.3
Jiang et al, 2018[15]	–	86.6	82.5	90.5	–	–	–	–	–	–	84.1	80	88.1
Pooled estimates	71.2	73.2	76.8	75.8	25.2	35	16.4	21.6	20.8	18.3	79.8	75	85.4

Abbreviations: DP, duraplasty; DS, dura-splitting.

Table 8 Summary of complication rates

Author, year	Overall (%)			CSF related[a] (%)			Infection (%)			Neurological deficits (%)			Mortality (%)		
	All	DS	DP	All	DS	DP	All	DS	DP	All	DS	DP	All	DS	DP
Limonadi and Selden, 2004[8]	4	—	8	—	0	—	4	0	8	—	—	—	—	—	—
Navarro et al, 2004[9]	14.6	3.5	26.4	13.7	1.7	26.4	1.8	1.7	1.8	0.9	0	1.8	—	—	—
Galarza et al, 2007[10]	8.3	—	—	5	—	—	—	—	—	1.6	—	—	—	—	—
Litvack et al, 2013[11]	7	3	13	0.9	0	2.1	6.3	3.2	10.6	—	—	—	—	—	—
Pomeraniec et al, 2016[12]	4	0	9	—	—	—	—	—	—	4	0	9	0	0	0
Pisapia et al, 2017[14]	—	—	—	8.4	0	17.5	2.1	3	1	—	—	—	1.5	2	1
Grahovac et al, 2018[13]	6.2	0	16.6	6.2	0	16.6	0	0	0	0	0	0	0	0	0
Jiang et al, 2018[15]	35.3	12.5	57.1	20.7	2.5	38.1	7.3	5	9.5	—	—	—	—	—	—
Pooled estimates	14.3	4.9	27.4	9.3	0.7	20	3.7	2.8	4.7	1.4	0	2.8	1.3	1.6	0.9

Abbreviations: CSF, cerebrospinal fluid; DP, duraplasty; DS, dura-splitting.
[a]Including CSF leak, pseudomeningocele, and aseptic meningitis.

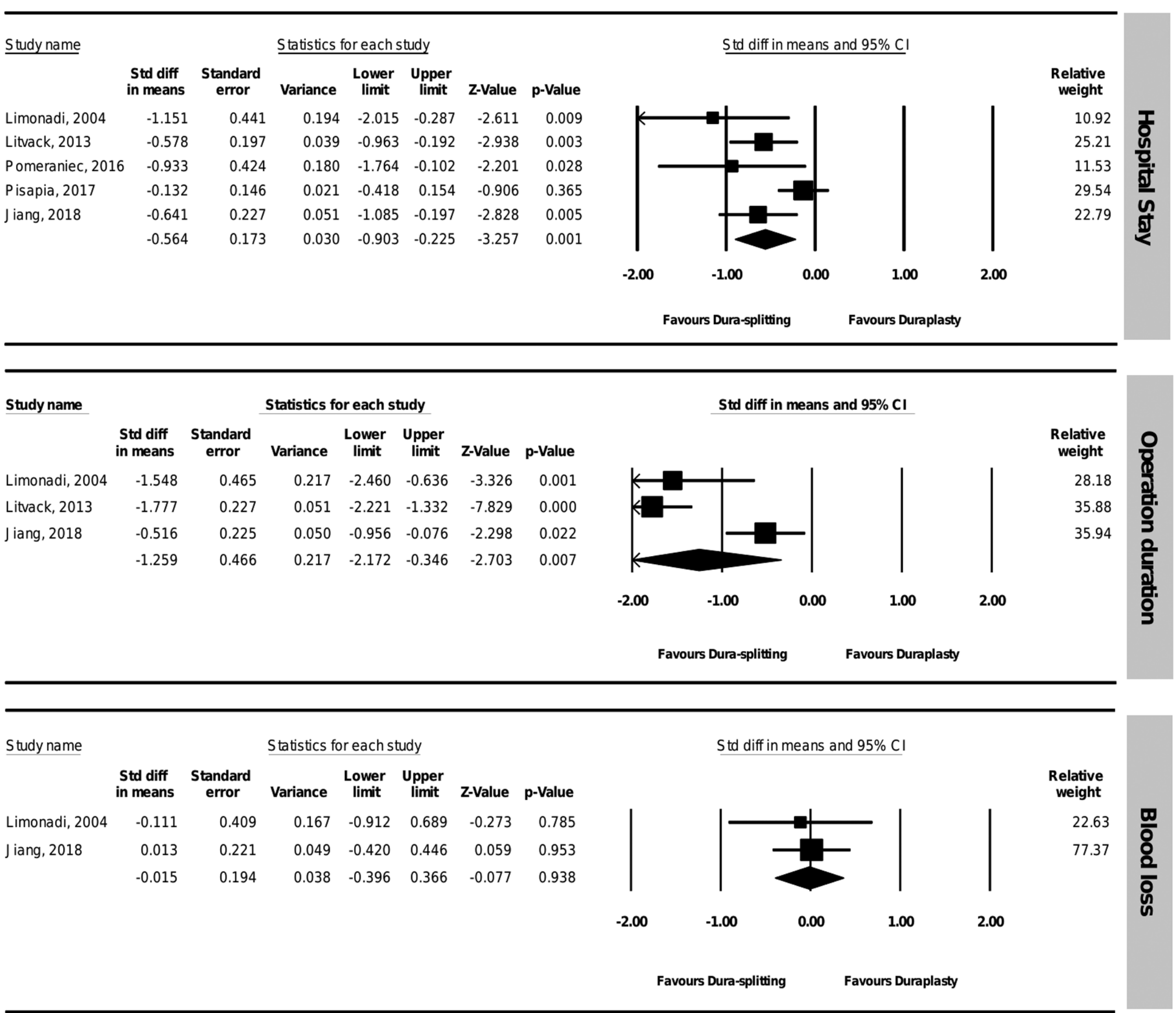

Fig. 3 Forest plots presenting effect sizes and their 95% confidence intervals for operative parameters.

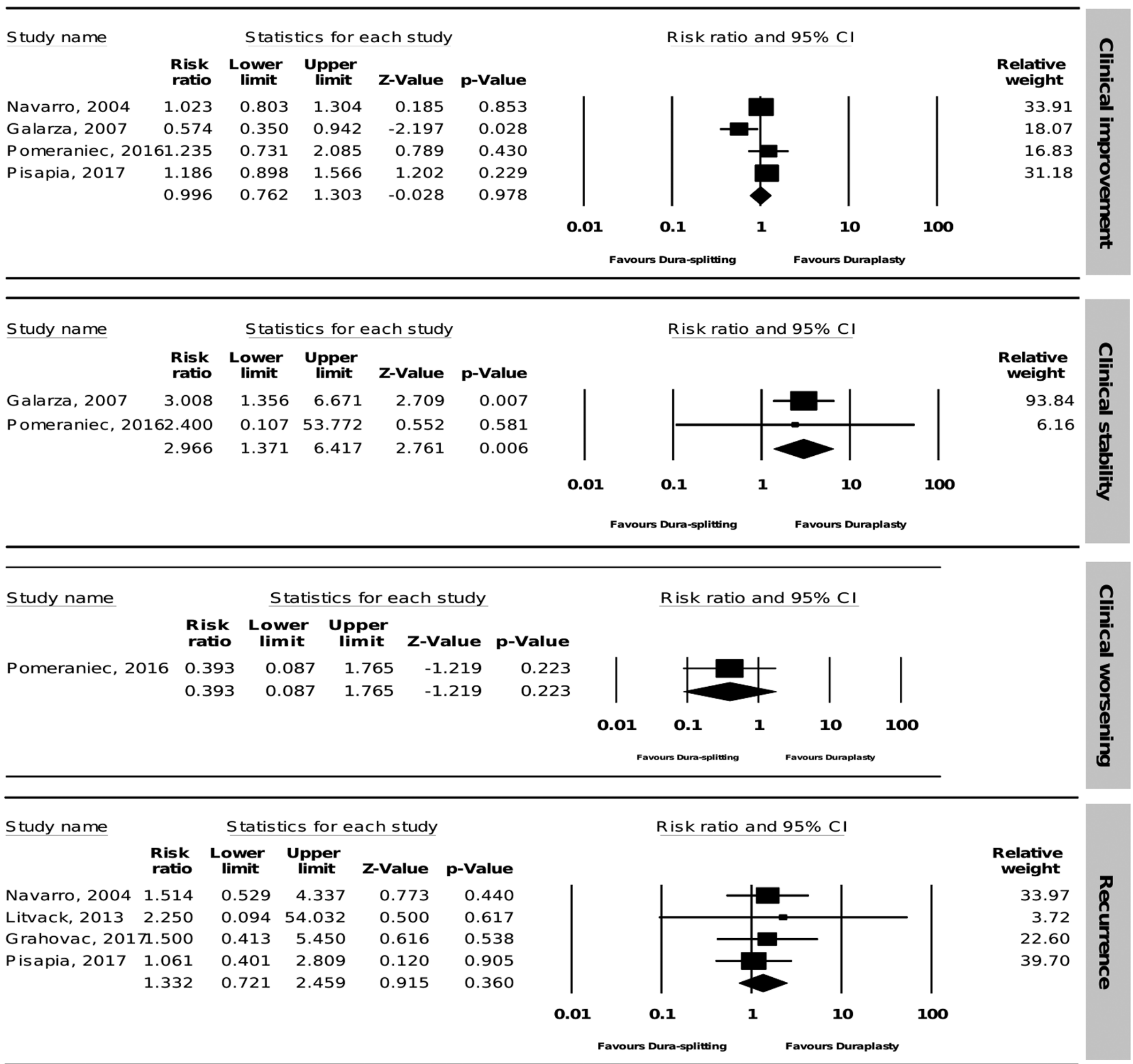

Fig. 4 Forest plots presenting effect sizes and their 95% confidence intervals for clinical outcome measures.

expected that the dura-splitting technique can result in a higher rate of postoperative syrinx resolution compared to foramen magnum decompression alone.

Operative Parameters

The shorter operation duration of the dura-splitting technique, which was expected as a result of sparing duraplasty as a time-consuming step, highlights the superiority of this technique over duraplasty in patients with coexisting medical morbidities who will benefit from a shorter procedure. Shorter operation duration also has the advantage of a lower postoperative infection rate. Interestingly, our meta-analysis has failed to show the superiority of the less invasive dura-splitting technique in terms of intraoperative blood loss. But, the dura-splitting technique has shown the advantage of significantly less intraoperative bleeding in the adult population.[3] These paradoxical results can be due to more rigorous hemostatic measures taken by neurosurgeons while operating on pediatric patients. The length of hospital stay was significantly shorter in the dura-splitting cohort. Hospital stay length is a significant factor mirroring various other factors such as the patient clinical condition or the occurrence of complications and also is an important variable for the institutional health-related policymakers to estimate the cost-effectiveness of a specific therapeutic modality. Therefore, the results of this review can be interesting for both neurosurgeons and policymakers.

Clinical Outcome

The most common postoperative clinical scenario was significant clinical improvement regardless of the surgical technique, followed by clinical stability as the second common outcome. The lack of any significant difference between the clinical outcome of patients in the two cohorts of our review challenges the opponent's proposals that the dura-splitting technique may not be adequately efficient to clinically improve patient symptoms.

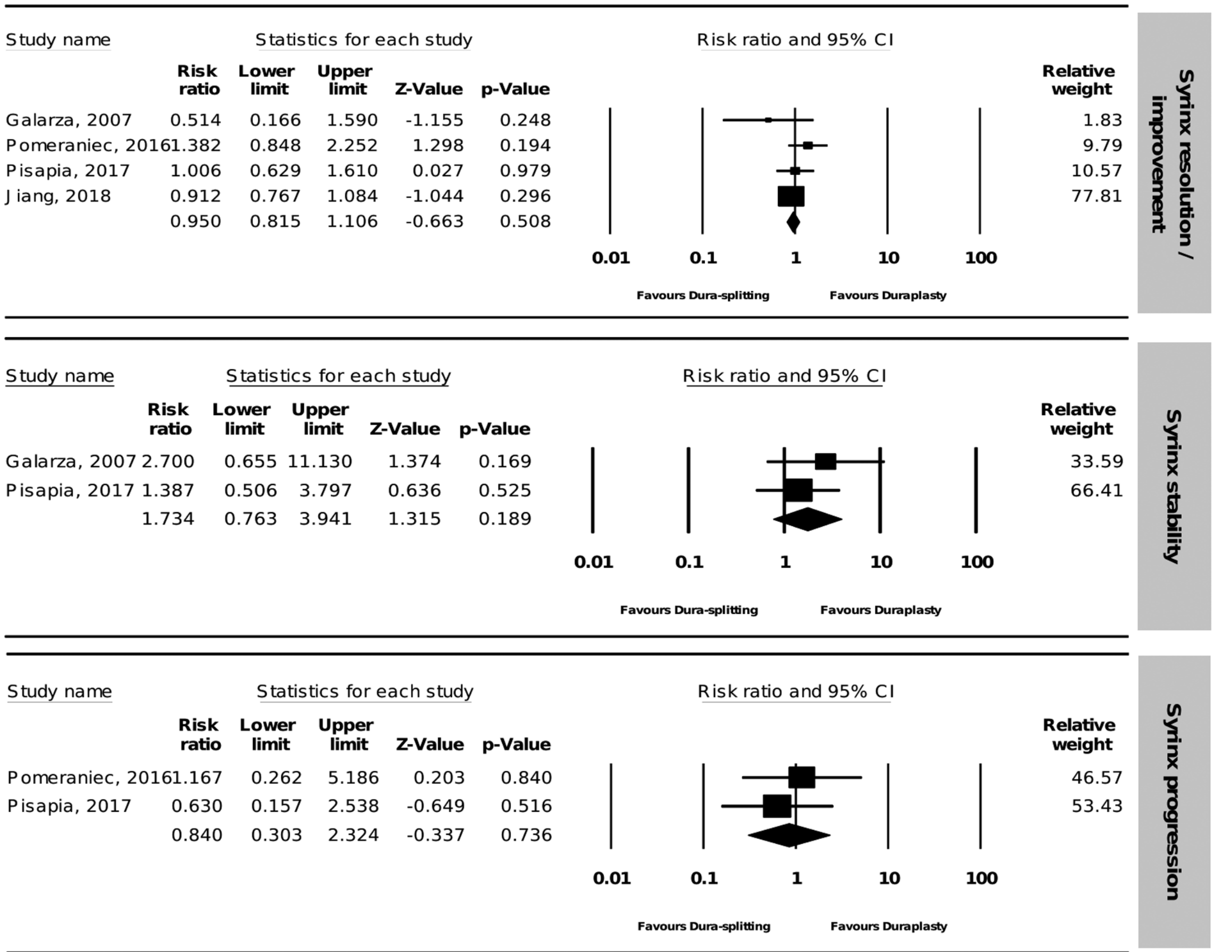

Fig. 5 Forest plots presenting effect sizes and their 95% confidence intervals for radiological outcome measures.

Although there was a significantly higher rate of clinical stability in the dura-splitting group, this deduction was made based on available data from two included studies and its significance may be biased as a result of the limited sample size. Generally, it can be suggested that a nearly similar clinical outcome can be anticipated following both the dura-splitting and duraplasty techniques. However, the included studies may have different definitions for outcome measures such as clinical improvement and stability, given the fact that a universally accepted clinical outcome rating scale is not yet available and this seems to be a blind spot that should be addressed.

It seems that there is a significant trend among neurosurgeons toward the implementation of the duraplasty technique for patients with a preoperative syrinx. This was also evident in our review cohort with a significantly higher rate of preoperative syrinx in the duraplasty group ($p < 0.001$). So, we decided to investigate whether the similarity of clinical outcomes between the two review cohorts is due to the confounding effect of less preoperative syrinx rate in the dura-splitting group or not. Unfortunately, only one of the included studies provided the necessary individual patient data to investigate this confounding effect.[12] After performing a logistic regression analysis on the available data from that study, the presence of preoperative syrinx did not show a significant effect on the equation and its potential confounding effect was not confirmed ($p > 0.05$). However, this analysis was performed on a relatively small sample size and its results should be interpreted cautiously.

The wide range of follow-up duration mentioned in the included studies may raise the concern that the studies with shorter follow-up duration may have missed important outcome data such as clinical and/or radiological improvements or worsenings. However, the approximate similarity of follow-up duration between the two review cohorts alleviates this concern to some extent.

As can be seen, the pooled estimate for the recurrence/reoperation rate did not significantly differ between the two surgical techniques. Recently, these rates were reported as 10 and 2% in the adult population, respectively, with no significant difference after the meta-analysis of data.[3] This highlights the dura-splitting technique as an efficient surgical option for the management of CM-I malformation in both pediatric and adult populations.

Radiological Outcome

The similarity of the two review cohorts in terms of radiological outcome was an interesting and hallmark finding of our review. The most common fate of syringomyelia following both surgical techniques was complete syrinx resolution. The similar findings of our recent systematic review, aimed

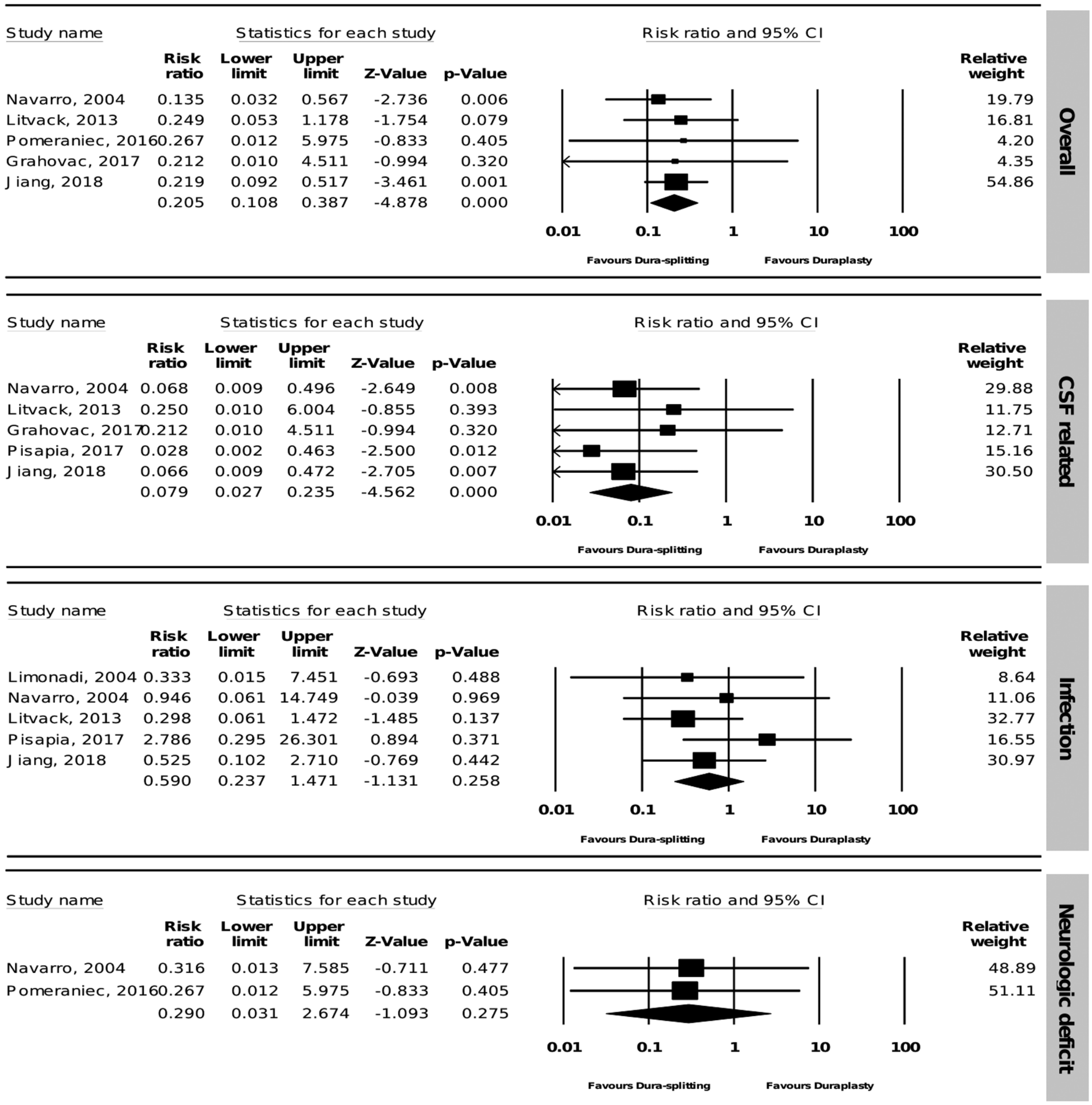

Fig. 6 Forest plots presenting effect sizes and their 95% confidence intervals for complication rates.

at demonstrating the outcomes of the dura-splitting technique in adult CM-I patients, have significantly alleviated the concern about the efficacy of the dura-splitting technique in the radiological improvement of adult patients with coexisting syringomyelia.[3] In the present review, we have reached similar results after a meta-analysis of data derived from the pediatric population. Preoperative syrinx size may play as a predicting variable for the postoperative rate of syrinx resolution or size reduction following these surgical techniques, rendering a larger syrinx less amenable to resolution following the dura-splitting technique. Among four studies with available data about radiological outcomes following both surgical techniques,[10,12,14,15] only two studies included simultaneous data related to the syrinx size[12,15] and only one of these two studies provided the individual patient data regarding preoperative syrinx size and postoperative syrinx course.[12] We decided to perform a reanalysis of the available individual patient data to investigate this potential correlation between preoperative syrinx size and postoperative syrinx improvement and there was no significant correlation in neither dura-splitting nor duraplasty groups ($p > 0.05$).

Hydrodynamic theory and craniospinal CSF pressure dissociation theory are among the pathophysiological theories trying to elucidate the mechanism responsible for syrinx formation in CM-I.[23,24] In a biomechanical study performed on craniocervical junction dura to quantify and compare mechanical properties of full dura and split dura, Chauvet et al showed that the inner layer of dura has different mechanical properties from the outer layer. This study revealed the high capability of the inner dural layer for expansion and concluded that splitting of the dura and elimination of the outer constraining dural layer can lead to posterior fossa expansion in Chiari patients.[25] This eventual dural expansion can explain the resolution of the syrinx in the dura-splitting technique by the elimination of the pathophysiological cause. These findings are intraoperatively demonstrated using ultrasonographic CSF flow measurement during the dura-splitting technique.[26]

Complications

Postoperative complications, mainly CSF-related and infection, are traditionally a significant concern of neurosurgeons who manage patients with Chiari malformation. CSF leakage is the most common complication after surgical techniques that include dura opening.[27] Significantly higher CSF-related complication rates of the duraplasty technique, which is confirmed by the results of our review, were the main provoking factor for the introduction of less invasive techniques that avoid the dura opening step. Based on the results of this review, the dura-splitting technique has the significant advantage of significantly fewer CSF-related complications. However, there are very few reports of CSF-related complications following the dura-splitting technique probably due to the inadvertent breach in the inner dural layer during the splitting process.[28] Infection in form of surgical site infection or meningitis is another serious complication more expected in the duraplasty technique. However, both techniques have been shown to have a similar infection rate in our review and this finding is in contrast to the reported higher infection rate following the duraplasty technique in the management of adult CM-I malformation. This discrepancy shows that variables other than the extent of invasiveness of the selected approach or the operation duration are involved in the pathophysiology of postoperative infectious complications in the pediatric population.

Strength and Limitations

So far, this is the first systematic review exclusively performed on the outcomes of the dura-splitting technique in pediatric patients with CM-I. We performed this review based on a previously designed and registered review protocol to ensure the quality of the review and commitment to a strict methodology. We have also prepared this review based on the MOOSE guideline that is a widely accepted guideline to prepare systematic reviews that include observational studies. The critical phases of review such as screening of the articles, assessing the quality of included studies, and data extraction was handled by two authors independently to reduce error and bias. The considerably long follow-up duration of both review cohorts compared to other reviews in the field of CM-I augments the reliability of our long-term outcome estimates.[29] This ensures more reliable estimates of long-term outcomes. Also, the quality of included studies was assessed as fair for the vast majority of included studies that is a stronghold for this review. The retrospective and observational nature and small sample sizes of included studies were among the limitations of our review. We have also faced some limitations in the process of subgroup analysis because the majority of included studies did not provide the required individual patient data and we also could not gain access to the data by contacting the corresponding authors.

Conclusion

It seems that the clinical and radiological outcomes following the surgical management of pediatric CM-I patients using dura-splitting and duraplasty techniques are comparable to each other. Furthermore, the dura-splitting technique has a significant advantage over duraplasty in terms of operation duration, hospital stay, and complication rates. Although this review provides the highest level of evidence available in the literature so far on this subject, our conclusions are derived from the data extracted mostly from retrospective studies. Therefore, future large-scale prospective studies can have a significant role in validating our results.

Authors' Contributions

Amin Tavallaii conceptualized the study. Amin Tavallaii, Ehsan Keykhosravi, and Ahmad Ghorbanpour were involved in article screening. Amin Tavallaii, Ahmad Ghorbanpour, and Ali Shahriari helped in data extraction. Amin Tavallaii, Ehsan Keykhosravi, and Hamid Rezaee helped in analysis. Amin Tavallaii and Hamid Rezaee drafted the work. All authors provided final approval.

Conflicts of Interest

None to declare.

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