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Efficacy of acupotomy for cerebral palsy
A systematic review and meta-analysis

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Abstract

Background: In children, cerebral palsy (CP) is one of the most common causes of irreversible neurological sequelae. Acupotomy, a modernized acupuncture form combining the effects of microsurgery and conventional acupuncture, may show specific benefits in the treatment of CP, especially with respect to spasticity. The aim of this review was to evaluate the efficacy of acupotomy for CP.

Methods: Eleven databases were comprehensively searched from their inception dates to November 27, 2018. Randomized controlled trials (RCTs) or quasi-RCTs evaluating acupotomy as a monotherapy or as adjunctive therapy to rehabilitation treatment for CP were included. The methodological quality of included studies was assessed using the risk of bias tool. The quality of evidence for each main outcome was evaluated using the Grading of Recommendations Assessment, Development, and Evaluation approach. Meta-analysis was performed, and the pooled data were presented as mean difference (MD) with 95% confidence interval (CI) for continuous outcomes and as risk ratio (RR) with 95% CI for dichotomous outcomes.

Results: Eight studies involving 530 participants were included. In 1 study, acupotomy was associated with significantly higher total effective rate (TER) compared with Bobath ($P < .01$). Acupotomy combined with rehabilitation was associated with significantly higher TER (RR 1.24, 95% CI 1.01–1.52, $I^2 = 77\%$) and gross motor function measure score (MD 12.62, 95% CI 11.75–13.49, $I^2 = 54\%$), and significantly lower muscle tone of gastrocnemius measured by the Ashworth scale or the modified Ashworth scale (MD −0.97, 95% CI −1.07 to −0.88, $I^2 = 0\%$) compared with rehabilitation alone. No studies reported the incidence of adverse events. The methodological quality of the included studies and quality of evidence for the main finding were generally low.

Conclusion: Current evidence shows that acupotomy as a monotherapy or as adjunctive therapy to rehabilitation treatment might have benefits in the treatment of CP. However, due to the small number of studies included, the lack of sample size, poor methodological qualities, and low quality of evidence, the findings of this review should be interpreted with caution. Larger and more rigorous, high-quality RCTs should be performed on this topic.

PROSPERO registration number: CRD42018105891.

Abbreviations: AS = Ashworth scale, CI = confidence interval, CP = cerebral palsy, FIM = functional independence measure, GAS = goal attainment scaling, GMFM = gross motor function measure, ICF = international classification of functioning, disability, and health, MAS = modified Ashworth scale, MD = mean difference, PEDI = pediatric evaluation of disability inventory, RCTs = randomized controlled trials, ROM = range of motion, RR = risk ratio, TER = total effective rate.

Keywords: acupotomy, cerebral palsy, meta-analysis, systematic review

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1. Introduction

Cerebral palsy (CP) is one of the most common causes resulting in irreversible neurological sequelae in children; it comprises a heterogeneous group of conditions including permanent nonprogressive central motor dysfunction affecting muscle tone, posture, and movement.[1] The overall prevalence of this disorder was known to be 2.11 per 1000 live births.[2]

The causes of CP are multifactorial, and prenatal factors such as prematurity and/or low birth weight have been reported to be most associated with an increased risk of CP.[3] Classification of CP is based on the type and distribution of motor abnormalities, which is noticeable after 18 to 24 months.[4] Based on children who were 8 years old and living in areas of Alabama, Georgia, Missouri, and Wisconsin in 2008, the prevalence of spastic CP was the highest (77.4%).[5]

Currently, there is no fundamental cure for the brain insults leading to motor dysfunction and no specific drug treatment for CP. The current therapy for CP includes various kinds of functional rehabilitation training aiming to help people with the condition lead an independent life as much as possible.[6] However, a recent review concluded that Bobath therapy, which is popular for the treatment of CP was unsatisfactory because

there was strong evidence for the lack of improvement of contracture and tone and weak evidence for the lack of improvement in function.[7] Therefore, new, safe and effective treatments are needed for CP.

Acupuncture is one of the most popular complementary and alternative therapies. It has been reported to suppress inflammation,[8] oxidative stress,[9] and neuronal apoptosis,[10] and improve neurobehavioral ability in animal models of CP. Clinical studies have also shown that acupuncture improves motor function,[11] alleviates adductor muscle tension,[12] and promotes the development of intelligence[13,14] in children with CP; therefore, it seems to be promising for the treatment of CP.[15]

Acupotomy is a kind of modern style acupuncture treatment, which uses a blade needle combined with a flat surgical scalpel at the tip of the needle (Fig. 1).[16] Recent studies have shown that acupotomy is clinically effective in the treatment of several musculoskeletal pain conditions.[17–21] It is thought that this novel treatment combines the effects of microsurgery and conventional acupuncture; therefore, in addition to the effects of acupuncture for CP, acupotomy may show specific benefits in the treatment of CP, especially in the spastic symptoms. Although the origins of the spasticity are in brain, the induced spasticity of the local muscles and joints can cause a vicious circle, limiting the movements and consequentially aggravating the disability.[22] Therefore, the use of a needle-tipped flat surgical scalpel to incise soft tissue can be used specifically to improve the spasticity and contracture in CP.

Recently, randomized controlled trials (RCTs) have been carried out on the use of acupotomy for CP, but the possibility of its use has not yet been systematically reviewed. Therefore, the aim of this review was to evaluate the efficacy and safety of acupotomy for CP based on the literature published so far.

2. Methods

2.1. Study registration

The protocol for this study was registered in the International Prospective Register of Systematic Reviews, PROSPERO: CRD42018105891 (URL: http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42018105891).

2.2. Search strategy

The initial search date was July 21, 2018, and that of the update search was November 27, 2018. The following English, Chinese, and Korean databases were comprehensively searched from their inception dates to search date: Medline (via PubMed), EMBASE (via Elsevier), the Cochrane Central Register of Controlled Trials, Allied and Complementary Medicine Database (via EBSCO), Cumulative Index to Nursing and Allied Health Literature (via EBSCO), China National Knowledge Infrastructure, Wanfang Data, VIP, Oriental Medicine Advanced Searching Integrated System, Research Information Service System, and Korea Citation Index. We reviewed the reference lists of relevant articles to identify additional trials. The following search terms were used in Medline: (acupotomy OR acupotomology OR acupotome OR "needle knife" OR "needle scalpel" OR "miniscalpel" OR "stiletto needle" OR "sword-like needle" OR "mini needle knife" OR "xiaozhendai") AND ("cerebral palsy" [MeSH Terms] OR "cerebral palsy" OR "cerebral paralysis"). The search strategy used in other databases is provided in Supplementary Digital Content 1, http://links.lww.com/MD/C770.

2.3. Study selection

The study selection criteria of this review were as follows:

(1) Types of studies: RCTs and quasi-RCTs that used quasi-random methods such as alternate allocation or allocation by birth date.
(2) Types of participants: Studies involving participants with a diagnosis of CP. There were no limitations of age, sex, or race. We excluded trials that included participants suffering from other serious illness such as cancer, liver disease, or kidney disease.
(3) Types of interventions: Studies using acupotomy or acupotomy combined with rehabilitation as experimental interventions, while using rehabilitation alone as controls. In this review, rehabilitation treatment was defined as a nonpharmacological approach used for CP including physical therapy, exercise therapy, occupational therapy, language therapy, casts, and so on, which is currently main treatment for CP.
(4) Types of outcomes: The primary outcomes were the spasticity measured using Ashworth scale (AS) or the modified Ashworth scale (MAS), and the improvements of the range of motion (ROM) of affected joints including ankle, knee, or hip, and the motor skills measured by such as the gross motor function measure (GMFM). The secondary outcomes were self-care ability measured using the pediatric evaluation of disability inventory (PEDI) or the functional independence measure (FIM), total effective rate (TER), and incidence of adverse events. There was no restriction based on language and publication type such as a journal article, dissertation,
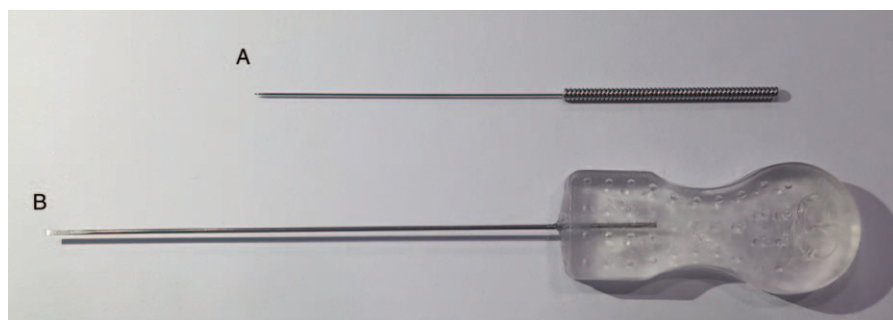
Figure 1. Image of acupotomy needle. A: filiform needle (Dongbang Medical Co, Korea); 0.25 mm × 30 mm. B: acupotomy needle (Dongbang Medical Co, Korea); 0.5 mm × 50 mm.

and conference proceeding. Two independent researchers (CY K and B L) carried out the database search and study selection, and any disagreement was resolved through discussion with other researchers.

2.4. Data extraction

Two independent researchers (CY K and B L) used predefined extraction forms to extract basic information of the included study and the data needed for meta-analysis. The extracted data included the information related to the risk of bias such as randomization method, allocation concealment, and blinding. Basic information of each included study such as the publication year, sample size, details of participants and interventions, details of acupotomy methods, treatment period, frequency of treatment, outcomes, and adverse events was also extracted. Any disagreement on data extraction were resolved through discussion with other researchers.

2.5. Quality assessment

Two researchers (CY K and B L) independently assessed the methodological quality of all included studies and the quality of evidence for each main finding. Any disagreement was solved through discussion with other researchers.

The methodological quality of included studies was assessed using the Cochrane Collaboration's risk of bias tool.[23] In this tool, seven items including random sequence generation, allocation concealment, blinding of participants and personnel, blinding of outcome assessment, completeness of outcome data, selective reporting, and other sources of bias are evaluated and categorized into "low risk," "unclear," or "high risk", to evaluate several risks of bias that can occur in RCTs. In the case of other sources of bias, it was evaluated as "low" if the characteristics of participants in each group were reported to be statistically homogeneous at baseline, but was otherwise rated "high". The results were presented as risk of bias graph and risk of bias summary using the Cochrane Collaboration's software program Review Manager (RevMan) version 5.3 for windows (Copenhagen, The Nordic Cochrane Centre, the Cochrane Collaboration, 2012).

We assessed the quality of evidence for main findings by using the grading of recommendations assessment, development, and evaluation (GRADE) approach.[24] Using the online program GRADEpro (https://gradepro.org/), we assessed the risk of bias; inconsistency, indirectness, and imprecision of the results; and the probability of publication bias using a 4-part scale ("very low," "low," "moderate," or "high").

2.6. Data analysis

Data analysis was conducted according to the type of comparison: acupotomy versus rehabilitation; and acupotomy combined with rehabilitation versus rehabilitation alone. Using RevMan version 5.3, the quantitative synthesis was performed on studies using the same comparison and outcome measure. The pooled data were presented as mean difference (MD) with 95% confidence interval (CI) for continuous outcomes and as risk ratio (RR) with 95% CI for dichotomous outcomes. Heterogeneity between the studies was assessed by the Chi-squared test and the I-squared statistic, and it was considered to be substantial when the I^2 value was 50% or more and considerable when it was 75% or more.

In the meta-analyses, the random effects model was used when the heterogeneity was significant (I-squared value ≥50%), while the fixed effects model was used when the heterogeneity was nonsignificant. The fixed-effect model was also used when the number of studies included in the meta-analysis was less than 5, where the estimates of inter-study variance had poor accuracy.[25,26] If the described data in the included study was insufficient or ambiguous, we contacted the corresponding author via email to request for additional information.

2.7. Subgroup analysis

The subgroup analysis was conducted according to the following:

(1) The type of primary data which used to calculate TER,
(2) The treatment period: short-term, less than 6-month; and long-term, 6-month or more,
(3) age of the participants, and
(4) follow-up period.

2.8. Sensitivity analysis

We conducted sensitivity analyses to prove the robustness of the meta-analysis result using exclusion criteria as follows:

(1) low-quality study in which less than 4 domains of the Cochrane group's risk of bias tool were evaluated as low risk of bias, and
(2) outliers that were numerically distant from the rest of the data.

2.9. Publication bias

If 10 or more studies were included in the meta-analysis, publication bias was assessed by using funnel plots.

3. Results

3.1. Description of studies

Among the 231 documents retrieved, 59 duplicate documents were excluded. After reviewing the titles and abstracts, 144 irrelevant documents were excluded, and full-texts of the other 28 were further reviewed. As results, 3 studies using same data from other articles (abstract proceedings), 3 case series, 4 review articles, and 10 studies using interventions incompatible with the study selection criteria of this review were removed. Therefore, a total of 8 studies[27–34] involving 530 participants were finally included in this systematic review, and 7 of them[27–32,34] were included in the meta-analysis (Fig. 2).

3.2. Characteristics of studies

All included studies were published in China from 2006 to 2017, and they were all journal articles. Of these, 6 studies were for spastic CP,[27–29,31,32,34] and the other 2 studies[30,33] did not mention the type of CP in the participants. The age of participants ranged from 1 to 16 years, and there were no adult participants. There were 6 parallel RCTs comparing acupotomy combined with rehabilitation versus rehabilitation alone.[28–30,32–34] The other 2 were parallel RCTs comparing 3 groups. These include acupotomy versus rehabilitation versus acupotomy combined with rehabilitation[27]; or acupuncture versus rehabilitation versus acupotomy combined with rehabilitation,[31] respectively.
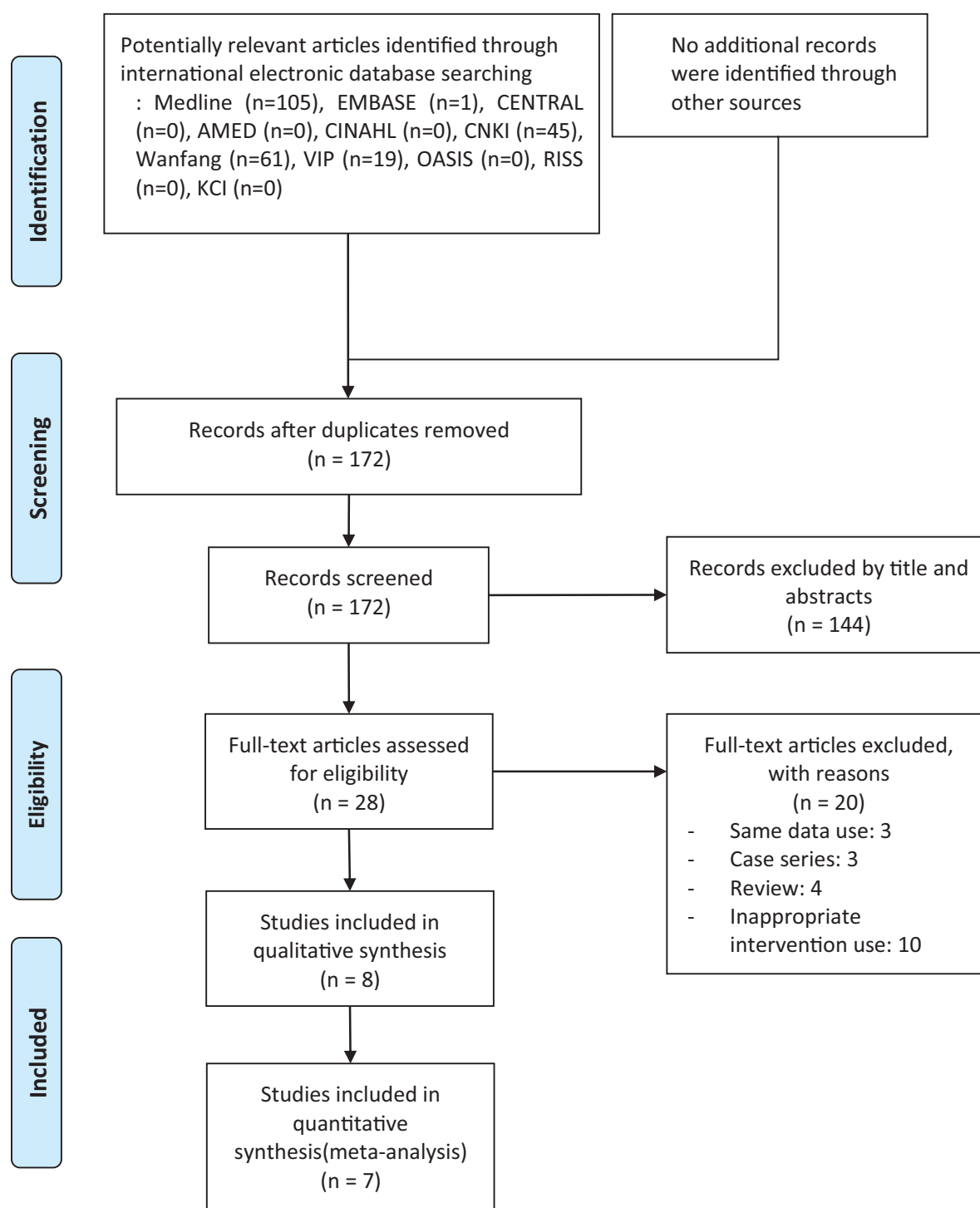
From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(6): e1000097. doi:10.1371/journal.pmed1000097*

For more information, visit www.prisma-statement.org.

Figure 2. PRISMA flow chart. PRISMA = preferred reporting items for systematic reviews and meta-analyses.

The treatment period ranged from 14 days to 6 months, with a median of 3 months. Most of the studies, except for 2,[29,34] were using treatment within 3 months. The most-frequently-used outcomes were TER in 5 RCTs.[27,28,30,32,34] However, in the 2 studies,[27,32] the TER was determined by improvement in MAS score, while in the other 3, the TER was determined by improvement in clinical symptoms,[30] clinical symptoms and MAS scores,[28] or clinical symptoms, MAS and GMFM

Table 1

Characteristics of included studies.

Author, yr	Sample size (G1:G2:G3)	Mean age (range)	Condition	G1 intervention	G2 intervention	G3 intervention	Duration	Frequency	Outcomes
Yan (2006)[27]	105 (35:34:36)	NR (2–4 yr)	Spastic cerebral palsy	Acupotomy	Bobath	Acupotomy plus Bobath	1 mo	Acupotomy: 2 sessions (1, 11 da) Bobath: 45 min/1 session/da Note. For G3, Bobath was performed from the third day after acupotomy.	TER (by MAS score) at postintervention
Zhao (2013)[28]	90 (45:45)	G1: 36.6±7.2 mo (24–48 mo) G2: 36.5±8.4 mo (18–46 mo)	Spastic cerebral palsy	Acupotomy plus rehabilitation	Rehabilitation	–	3 mo	Acupotomy: 1 session/mo Rehabilitation: NR	1) MAS score (the target muscle was not described) at postintervention 2) GMFM score at postintervention 3) TER (by clinical symptoms, MAS) at postintervention
Liang et al (2014)[29]	54 (27:27)	G1: 46.7±8.1 mo (NR) G2: 47.4±7.6 mo (NR)	Spastic cerebral palsy	Acupotomy plus rehabilitation	Rehabilitation	–	6 mo	Acupotomy: 1 session/mo Rehabilitation: NR Note. For G1, rehabilitation was performed from the third day after acupotomy.	1) AS score (Gastrocnemius) (1) at post-intervention (2) at 1 yr f/u 2) Foot dorsiflexion angle (passive) (1) at postintervention (2) at 1 yr f/u 3) GMFM score (1) at postintervention (2) at 1 yr f/u
Xue (2015)[30]	56 (28:28)	G1: 22±7 mo (12–36 mo) G2: 21±9 mo (12–36 mo)	Cerebral palsy	Acupotomy plus casts	Casts	–	2 wk	Acupotomy: single session Casts: keeping for 14 da	TER (by clinical symptoms) at postintervention
Chen (2016)[31]	30 (10:10:10)	NR (3–12 yr)	Spastic cerebral palsy	Acupuncture	Bobath	Acupotomy plus Bobath	3 mo	Acupuncture: 1 session/da Bobath: 30 min/1 session/da *Acupotomy: 1 session/2 wk *Note. For G3, Bobath was performed from the third day after acupotomy.	1) Flexion angle (active) of ankle, knee, and hip joint at post-intervention 2) MAS score (soleus, gastrocnemius, quadriceps femoris) at postintervention 3) Locomotion score (walking or wheelchair) in FIM at postintervention
Ye (2016)[32]	54 (30:24)	NR (4–12 yr)	Spastic cerebral palsy	Acupotomy plus Bobath	Bobath	–	3 mo	Acupotomy: 1 session/wk (≥5 yr old), 1 session/2 wk (<5 yr old) Bobath: 4 5min/1 session/da	TER (by MAS score) at postintervention
Naerbuli (2017)[33]	101 (51:50)	G1: 8±1 yr (1–16 yr) G2: 9±1 yr (2–16 yr)	Cerebral palsy	Acupotomy plus Bobath, Rood, Vojta, Ueda, occupational therapy, speech therapy, psychological rehabilitation, conductive education	Bobath, Rood, Vojta, Ueda, occupational therapy, speech therapy, psychological rehabilitation, conductive education	–	3 mo	Acupotomy: 1 session/mo Rehabilitation: NR	1) Footprint ratio at post-intervention 2) Weight-bearing X-ray at postintervention
Tan (2017)[34]	40 (20:20)	G1: 6.9±0.5 yr (1–15 yr) G2: 6.4±0.7 yr (1–13 yr)	Spastic cerebral palsy	Acupotomy plus rehabilitation	Rehabilitation	–	6 mo	Acupotomy: 1 session/mo Rehabilitation: NR	1) MAS* score (gastrocnemius) at postintervention 2) GMFM score at postintervention 3) TER (by clinical symptoms, MAS, GMFM score) at postintervention

AS=Ashworth scale, FIM=functional independence measure, GMFM=gross motor function measure, MAS=modified Ashworth scale, NR=not recorded, TER=total effective rate.
* The name of the scale was not described in the original. We tried to contact the corresponding author but could not, as there were no contacts left in the study. But this is strongly estimated as MAS.

scores,[34] respectively. In addition, the muscle tone of gastrocnemius measured by AS or MAS was reported in 3 studies,[29,31,34] the ROM of the joint contractures in 2,[29,31] the GMFM score in 3,[28,29,34] and the locomotion score in FIM in 1 study.[31] Meta-analysis was only possible for TER, the muscle tone of gastrocnemius measured by AS or MAS, and GMFM scores. No studies reported the incidence of adverse events (Table 1).

3.3. Risk of bias assessment

In random sequence generation, 3 studies[31,33,34] were assessed to have a low risk of bias which used a random number table, while the other 4 studies[27,29,30,32] were assessed to have a high risk of bias which used inadequate randomization method based on the order of treatment. One study[28] that did not describe the randomization method used considered to have an unclear risk of bias. No studies reported on allocation concealment. In blinding of participants and personnel, all included studies were assessed to have a high risk of bias due to the nature of the intervention. In blinding of outcome assessment, only 1 study[31] reported that the independent researcher assessed the outcomes, which was assessed to have a low risk of bias. No studies reported the dropout or withdrawal of participants. In selective reporting domain, 3 studies[27,30,32] were assessed to have a high risk of bias which used only TER, a secondary processed data, as their outcome. In other sources of bias, 6 studies[28–31,33,34] were assessed to have a low risk of bias as they reported no statistical differences in the characteristics of participants between the groups at baseline (Figs. 3 and 4).

3.4. Efficacy of acupotomy for CP

3.4.1. Acupotomy versus rehabilitation. There was 1 study comparing acupotomy and rehabilitation,[27] and only TER determined by improvement in MAS score at postintervention was used as an outcome. Bobath was used as the rehabilitation treatment in the treatment period of 1 month. According to the reported results, acupotomy was associated with significantly higher TER than Bobath ($P < .01$).

3.4.2. Acupotomy combined with rehabilitation versus rehabilitation alone. There were 7 studies comparing acupotomy combined with rehabilitation and rehabilitation alone.[28–34] Various rehabilitation treatment including Bobath or casts were used as the control interventions, and the mean treatment period was 14 weeks. According to the pooled results, the combined therapy was associated with significantly higher TER (RR, 1.24; 95% CI, 1.01–1.52; $I^2 = 77\%$) and GMFM score (MD, 12.62; 95% CI, 11.75–13.49; $I^2 = 54\%$), and significantly decreased muscle tone of gastrocnemius measured by AS or MAS (MD −0.97, 95% CI −1.07 to −0.88, $I^2 = 0\%$) compared with rehabilitation alone (Figs. 5–7) (Table 2).

In subgroup analysis according to the primary data of TER, however, only the result of TER determined by improvement in clinical symptoms and MAS score was of significance (RR 1.19, 95% CI 1.02–1.40). There were no significant differences on TER determined by improvement in MAS score (RR 1.65, 95% CI 0.45–6.07), in clinical symptoms (RR 1.08, 95% CI 0.93–1.25) or in clinical symptoms, MAS and GMFM scores (RR 1.29, 95% CI 0.93–1.77), between 2 groups. In subgroup analysis by treatment period, the results of long-term improvement in the muscle tone of the gastrocnemius measured using AS or MAS (MD, −0.98; 95% CI, −1.08 to −0.89; $I^2 = 0\%$) and improvement of the GMFM score at both short-term (MD, 13.14; 95% CI, 11.18–15.10) and long-term (MD, 12.49; 95% CI, 11.52–13.46; $I^2 = 75\%$) were significant. There were no significant differences in the results of both short-term (RR, 1.24; 95% CI, 0.97–1.58; $I^2 = 83\%$) and long-term TER (RR, 1.29; 95% CI, 0.93–1.77), and short-term improvement in the muscle tone of the gastrocnemius through AS or MAS (MD, −0.50, 95% CI, −1.29 to 0.29) between the combined therapy and rehabilitation alone groups (Table 2).

Sensitivity analysis performed by excluding low-quality studies showed that the superior effectiveness of the combined therapy group in improving the muscle tone of the gastrocnemius was no longer detected (MD, −0.50; 95% CI, −1.29 to 0.29); however, sensitivity analysis performed by excluding outliers showed that the combined therapy group consistently had superior effectiveness (MD, −0.98; 95% CI, −1.08 to −0.89; $I^2 = 0\%$). For
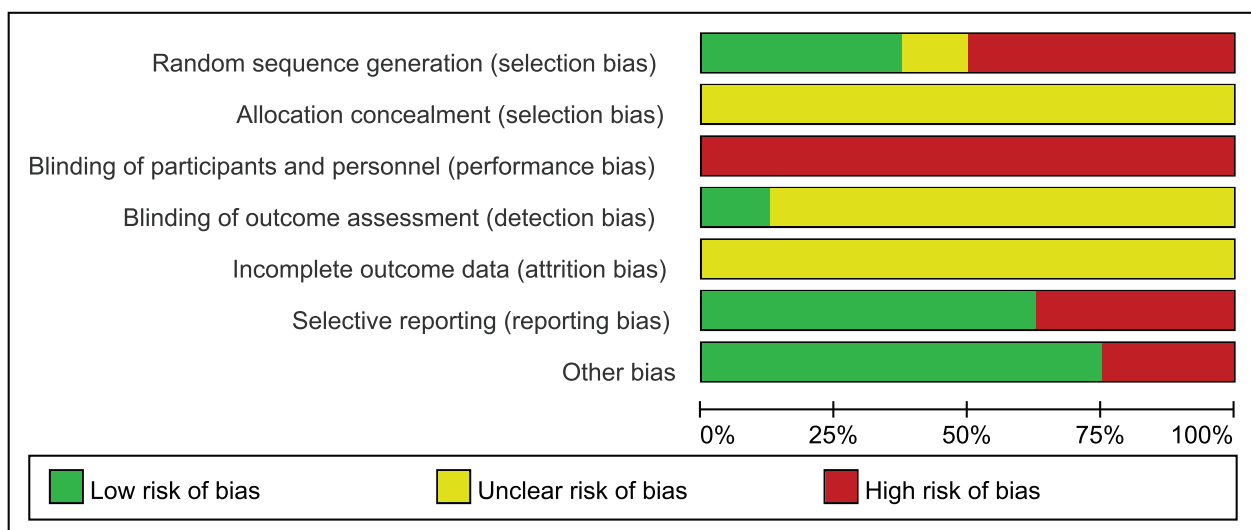

Figure 3. Risk of bias graph of included studies.

Figure 4. Risk of bias summary of included studies.

GMFM scores, sensitivity analysis performed by excluding outliers showed consistently significant efficacy in the combined therapy group compared with the rehabilitation alone group (MD, 12.49; 95% CI, 11.52–13.46; $I^2 = 75\%$) (Table 3).

Chen et al[31] reported that the muscle tones of soleus and quadriceps femoris measured by MAS of the combined therapy group were significantly improved when compared to the control group (both $P < .05$). The improvements in passive or active ROM of ankle, knee, or hip joints were reported in Liang et al[29] and Chen et al.[31] According to the reported results, the combined therapy group was associated with significantly superior results in the improvements in passive ROM of ankle dorsiflexion ($P < .05$),[29] and active ROMs of ankle flexion, knee flexion and hip flexion (all $P < .01$).[31] Moreover, Chen et al[31] reported that the combined therapy group was associated with significantly superior results in the locomotion score of FIM ($P < .05$). Naerbuli et al[33] reported the footprint ratios and weight-bearing lateral X-ray parameters of the combined therapy group were significantly improved when compared with the

control group at postintervention (all $P < .05$). Liang et al[29] conducted follow-up assessment after 1 year and reported significant improvement in the combined therapy group compared with the rehabilitation alone group in terms of the AS score ($P < .05$), foot dorsiflexion angle ($P < .01$), and GMFM score ($P < .05$).

3.5. Acupotomy methods for cerebral palsy

Among the included studies, there were only 4 studies[29–31,34] that referred to anesthesia before the acupotomy procedure. Chen et al[31] classified purpose of acupotomy procedure as cutting for correction, muscle stimulation, and nerve stimulation, but most of the other studies[27–30,32–34] have used acupotomy to incise the affected muscle or tendon and relevant acupoints. Especially, Achilles tendon[30,31,33] and gastrocnemius[26,32,34] were frequently reported as stimulation site. There was a lack of research to report procedure-related information such as texture or sensation indicating proper procedure, needle type, or qualifications of the practitioner. The total number of treatment sessions ranged from 1 to 12, and 6 sessions were most common. Treatment frequency was distributed from 1 session per week to 1 session per month, and it was most common to perform 1 session per month (Table 4).

3.6. Quality of evidence

In the comparison of acupotomy combined with rehabilitation and rehabilitation alone, the quality of evidence was graded as "very low" to "moderate" (Table 2). Especially, the quality of evidence for the improvement of the muscle tone of the gastrocnemius measured by AS or MAS and GMFM score were graded as "moderate"; therefore, we are moderately confident in the effect estimate. The main reasons for downgrading were the high risks of bias in the RCTs included in each meta-analysis, imprecision of the findings because of the small sample size and wide CIs, and indirectness of the outcome measure.

3.7. Publication bias

Because each meta-analysis contained fewer than 10 studies, the publication bias could not be evaluated.

4. Discussion

In the absence of a fundamental cure for CP, this study was conducted to evaluate the efficacy of acupotomy for CP, which is a new therapy combining microsurgery and conventional acupuncture, compared with rehabilitation treatment that is a typical first-line treatment for CP. Through a comprehensive search of English, Chinese, and Korean databases, a total of 8 studies[27–34] involving 530 participants were included in this review.

As a monotherapy, acupotomy showed significantly higher TER determined by improvement in MAS score compared with Bobath groups. As adjunctive therapy, acupotomy combined with rehabilitation had higher TER and GMFM score, and lower muscle tone of gastrocnemius, soleus, and quadriceps femoris compared with rehabilitation alone. In addition, combined therapy showed higher improvements in passive ROM of ankle dorsiflexion, active ROMs of ankle, knee, and hip flexion, locomotion score of FIM, the footprint ratios, and weight-

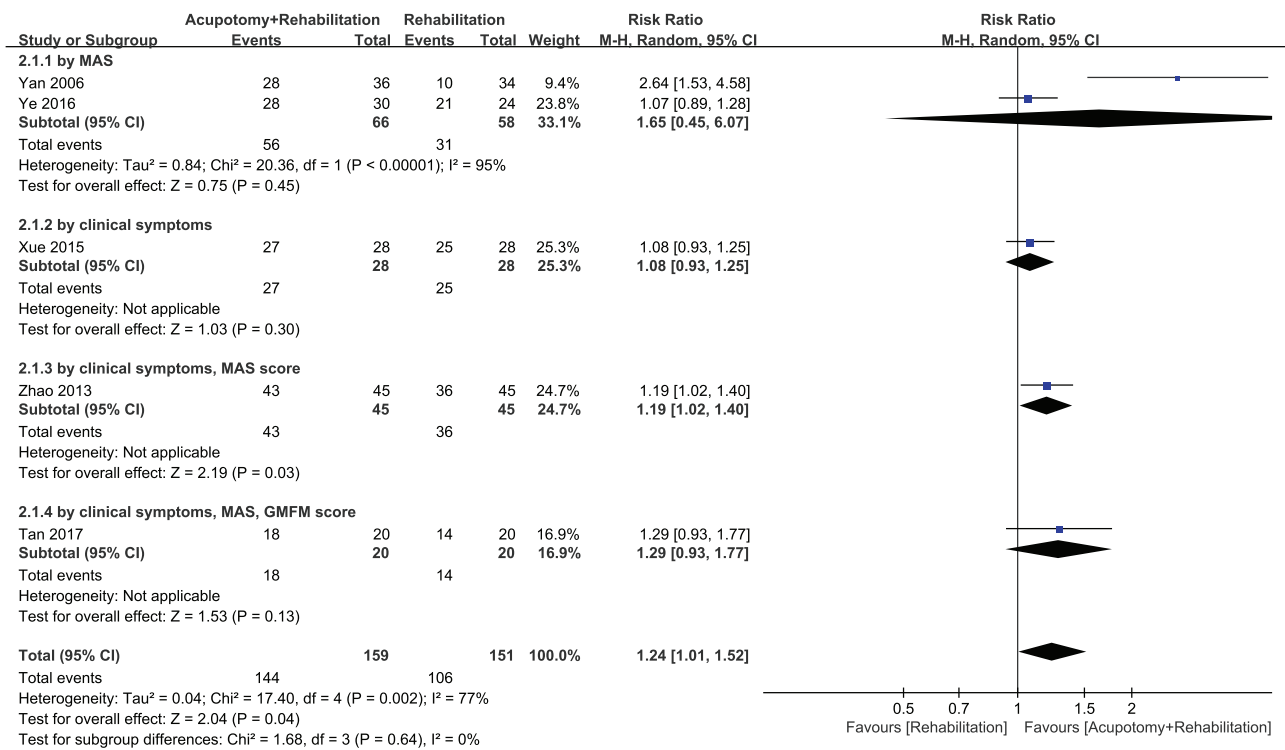**Figure 5.** Forest plots of total effective rate comparison: acupotomy combined with rehabilitation versus rehabilitation alone.
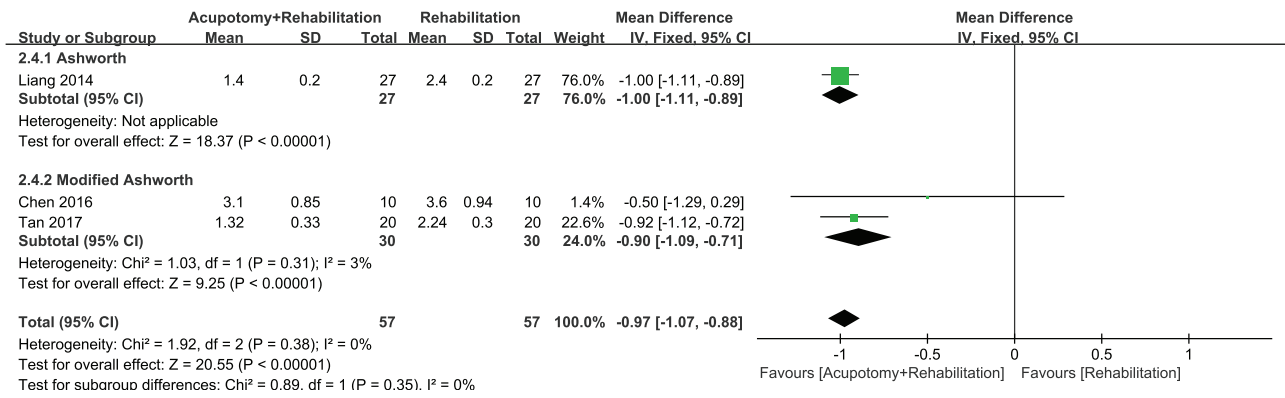

Figure 6. Forest plots of the Ashworth scale or modified Ashworth scale comparison: acupotomy combined with rehabilitation versus rehabilitation alone.
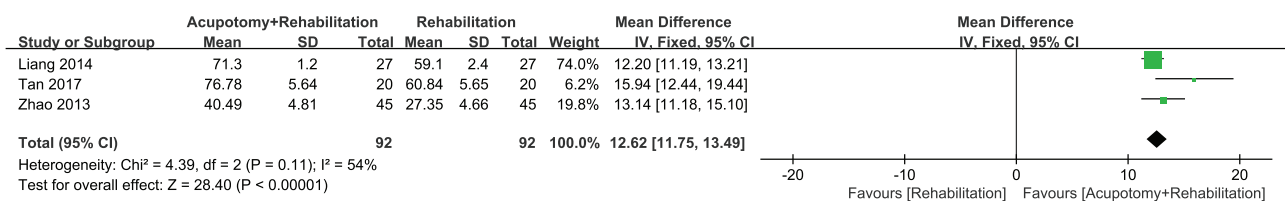

Figure 7. Forest plots of the gross motor function measure comparison: acupotomy combined with rehabilitation versus rehabilitation alone.

Table 2

Summary of findings: acupotomy plus rehabilitation compared with rehabilitation alone.

Outcomes		RCTs	Sample size	Anticipated absolute effects (95% CI)		Relative effect (95% CI)	Effects model	I^2 value	Quality of evidence (GRADE)	Comments
				Risk with rehabilitation	Risk with acupotomy plus rehabilitation					
TER	Total	5	310	702 per 1000	870 per 1000 (709–1000)	RR 1.24 (1.01–1.52)	Random	77%	⊕⊕◯◯ Low	Risk of bias (−1) Indirectness (−1)
	Subgroup set 1 by MAS score	2	124	534 per 1000	882 per 1000 (241–1000)	RR 1.65 (0.45–6.07)	Random	95%	⊕◯◯◯ Very low	Risk of bias (−1) Indirectness (−1) Imprecision (−1)
	By clinical symptoms	1	56	893 per 1000	964 per 1000 (830–1000)	RR 1.08 (0.93–1.25)	Random	NA	⊕◯◯◯ Very low	Risk of bias (−1) Indirectness (−1) Imprecision (−1)
	By clinical symptoms, MAS score	1	90	800 per 1000	952 per 1000 (816–1000)	RR 1.19 (1.02–1.40)	Random	NA	⊕◯◯◯ Very low	Risk of bias (−1) Indirectness (−1) Imprecision (−1)
	By clinical symptoms, MAS, GMFM score	1	40	700 per 1000	903 per 1000 (651–1000)	RR 1.29 (0.93–1.77)	Random	NA	⊕◯◯◯ Very low	Risk of bias (−1) Indirectness (−1) Imprecision (−1)
	Subgroup set 2 Short-term (<6 mo)	4	270	702 per 1000	871 per 1000 (681–1000)	RR 1.24 (0.97–1.58)	Random	83%	⊕⊕◯◯ Low	Risk of bias (−1) Indirectness (−1)
	Long-term (≥6 mo)	1	40	700 per 1000	903 per 1000 (651–1000)	RR 1.29 (0.93–1.77)	Random	NA	⊕◯◯◯ Very low	Risk of bias (−1) Indirectness (−1) Imprecision (−1)
AS or MAS score (gastrocnemius)	Total	3	114	–	MD 0.97 lower (1.07–0.88 lower)	–	Fixed	0%	⊕⊕⊕◯ Moderate	Risk of bias (−1)
	Subgroup set 1 By AS score	1	54	–	MD 1 lower (1.11–0.89 lower)	–	Fixed	NA	⊕⊕◯◯ Low	Risk of bias (−1) Imprecision (−1)
	By MAS score	2	60	–	MD 0.9 lower (1.09–0.71 lower)	–	Fixed	3%	⊕⊕◯◯ Low	Risk of bias (−1) Imprecision (−1)
	Subgroup set 2 Short-term (<6 mo)	1	20	–	MD 0.5 lower (1.29 lower to 0.29 higher)	–	Fixed	NA	⊕⊕◯◯ Low	Risk of bias (−1) Imprecision (−1)
	Long-term (≥6 mo)	2	94	–	MD 0.98 lower (1.08–0.89 lower)	–	Fixed	0%	⊕⊕◯◯ Low	Risk of bias (−1) Imprecision (−1)
GMFM score	Total	3	184	–	MD 12.62 higher (11.75–13.49 higher)	–	Fixed	54%	⊕⊕⊕◯ Moderate	Risk of bias (−1)
	Subgroup set 1 Short-term (<6 mo)	1	90	–	MD 13.14 higher (11.18–15.10 higher)	–	Fixed	NA	⊕⊕◯◯ Low	Risk of bias (−1) Imprecision (−1)
	Long-term (≥6 mo)	2	94	–	MD 12.49 higher (11.52–13.46 higher)	–	Fixed	75%	⊕⊕◯◯ Low	Risk of bias (−1) Imprecision (−1)

AS = Ashworth scale, CI = confidence interval, GMFM = gross motor function measure, GRADE = grading of recommendations assessment, development, and evaluation, MAS = modified Ashworth scale, MD = mean difference, NA = not applied, RCTs = randomized controlled trials, RR = risk ratio, TER = total effective rate.

bearing lateral X-ray parameters compared with rehabilitation alone.

Based on this study, we found that acupotomy as monotherapy or adjunctive therapy showed a consistently positive effect on CP. However, there are some concerns in interpreting the results of the review. There were 8 studies included in this review, and only 1 study[27] assessed the efficacy of acupotomy as a monotherapy. Moreover, the methodological quality of the included studies was generally low. Especially, only 3 studies[31,33,34] described appropriate randomization method and none reported concealment of allocation, resulting in high risk of selection bias; none of the studies performed blinding of the participants and personnel, and only 1 study[31] performed blinding of the outcome assessors. This may have resulted in the overestimation of the efficacy of

acupotomy for CP. In addition, the quality of evidence for main findings was generally low and there was no high-quality evidence. Therefore, owing to the small number of studies, small sample size, low methodological quality of included studies, and low quality of evidence for main findings, we could not draw firm conclusions about the efficacy of acupotomy.

We conducted a subgroup analysis of the efficacy comparing acupotomy plus rehabilitation and rehabilitation alone according to the primary data of TER and treatment period. There was a significant result in TER calculated by only clinical symptoms and MAS score. Notably, both the muscle tone of gastrocnemius measured by AS or MAS score showed significant results when acupotomy was conducted for long-term (≥6 months), but not when it was conducted for short-term (<6 months). This may

Table 3

Results of sensitivity analysis: acupotomy plus rehabilitation compared with rehabilitation alone.

Outcomes		RCT	Sample size	Effects model	MD	95% CI	I^2 value	Z value	P value
Ashworth scale score (gastrocnemius)	Excluding low quality studies	1	20	Fixed	−0.50	−1.29, 0.29	NA	1.25	1.21
	Excluding outliers	2	94	Fixed	−0.98	−1.08, −0.89	0	20.55	<.00001
GMFM score	Excluding outliers	2	94	Fixed	12.49	11.52, 13.46	75	25.18	<.00001

CI = confidence interval, GMFM = gross motor function measure, MD = mean difference, NA = not applied, RCT = randomized controlled trial.

Table 4

Acupotomy methods for cerebral palsy.

Author, yr	Anesthesia	Stimulation site	Texture or sensation indicating proper procedure	Needle type (needle diameter * length)	Total treatment session	Frequency and duration	Other intervention	Qualifications or experiences
Yan (2006)[27]	NR	Affected muscle (upper limbs and/or lower limbs)	NR	NR	2	2 sessions/mo, 1 mo	Bobath	NR
Zhao (2013)[28]	NR	6–10 acupoints –Upper limb spasticity: Shoulder 3 needles (Shoulder 3 needles), LI11, LI10 –Palmar flexion, thumb adduction: LU10, LU9, LI4, SI3 –Lower limb spasticity: GB30, BL36, BL40, BL57, ST36, GB34 –Plantar flexion: BL59, BL60, ST41	NR	Disposable needle knife (Beijing Huaxia Medical Instrument Factory)	3	1 session/mo, 3 mo	Rehabilitation	NR
Liang et al (2014)[29]	Local anesthesia with lidocaine	Gastrocnemius	Loose texture (practitioner)	NR	6	1 session/mo, 6 mo	Rehabilitation	NR
Xue (2015)[30]	Routine anesthesia	Achilles tendon	NR	NR	1	1 session/2 wk, 2 wk	Casts	NR
Chen (2016)[31]	Local anesthesia	[1] Cutting for correction: adductor magnus, long head of biceps femoris, semitendinosus, semimembranosus, hamstring, tendon sheath around the ankle, Achilles tendon, posterior tibial tendon, fibularis longus, fibularis brevis [2] Muscle stimulation: gluteus maximus, adductor magnus, long head of biceps femoris, semitendinosus, semimembranosus, quadriceps, tibialis anterior muscle [3] Nerve stimulation: spinal nerve (second lumbar vertebra level), peripheral nerve (popliteal fossa, lumbar plexus next to inguinal artery, sciatic nerve, femoral nerve, obturator nerve, common peroneal nerve)	[1] Bouncing sensation (practitioner) [2] Abnormal sensation (patients) [3] Radiation-like sensation (patients)	[1] No. 4 (NR) [2] No. 2 or 1 (NR) [3] No. 4 (NR)	6	1 session/2wk, 3 mo	Bobath	NR
Ye (2016)[32]	NR	Ligament around the ankle joint, gastrocnemius, soleus, soft tissue around the hip, adductor, gracilis, iliotibial tract, soft tissue around the spine	NR	NR	12 (≥5 yr old), 6 (<5 yr old)	1 session/wk (≥5 yr old), 1 session/2wk (<5 yr old), 3 mo	Bobath	NR
Naerbuli (2017)[33]	NR	Achilles tendon, posterior tibial tendon, peroneal tendon	NR	No. 4 (NR)	3	1 session/mo, 3 mo	Bobath, Rood, Vojta, Ueda, occupational therapy, speech therapy, psychological rehabilitation, conductive education	NR
Tan (2017)[34]	Local anesthesia with lidocaine	Gastrocnemius	Loose texture (practitioner)	NR	6	1session/mo, 6 mo	Rehabilitation	NR

NR = not recorded.

imply that a long enough period of acupotomy treatment is necessary for lowering muscle tone of gastrocnemius compared with rehabilitation alone. GMFM scores were significantly higher in the combined treatment group than the rehabilitation group for both the long-term and short-term treatment. Sensitivity analysis to identify the robustness of the meta-analysis results revealed the presence of a significant difference in the GMFM score but not the AS score; however, since only one or 2 studies were included in each subgroup, caution is required when interpreting the results of sensitivity analysis.

None of the literature included in this study evaluated the compliance of acupotomy and adverse events related to acupotomy. Invasive interventions can reduce the compliance of patient, especially in children,[35] and compliance is one of the important factors for the choice of treatment. Although acupuncture is considered to be safe,[36] acupotomy is more invasive than acupuncture and needs a more systematic evaluation of the safety for application.

The ultimate goal of treating patients with CP is to maximize potential benefit to patients and minimize disability, thereby enabling independent social life and improving the quality of life. In 2001, the World Health Organization published the International Classification of Functioning, Disability, and Health (ICF), and in assessing disability, it emphasized not only changes in the structure and function of the body following disease or accident, but also the social activities determined by interaction with the environment and the limitation of participation.[37] Parents, young people, and doctors have identified 8 domains for evaluating therapeutic effects in CP, comprising impairment, general health, gross motor skills, self-care/fine motor skills, speech/communication, integration/participation, quality of life, and caregiver issues according to ICF.[38] In addition, Novak et al[7] proposed outcome measures that sensitively evaluate the several domains. They suggested using the Modified Tardieu Scale for spasticity, the GMFM for gross motor skills, PEDI and activities scale for kids for self-care, the assisting hand assessment for fine motor, goal attainment scaling (GAS) for speech/communication, and GAS as well as the Canadian occupational performance measure for integration/participation.

There were no studies evaluating domains of speech/communication and integration/participation among the studies included in this review. In addition, there were only 2 studies[29,34] evaluating gross motor skills and only 1[31] evaluated self-care ability. Furthermore, in the 3 studies[27,30,32] included in this review, the efficacy of intervention was evaluated using only secondary processed TER without showing the raw data. This indirectness of outcome measures can be a risk of bias in assessing the efficacy of acupotomy for CP. Therefore, future studies should assess the efficacy of acupotomy using the sensitive outcome measures presented in previous studies.

Acupotomy is known to alleviate the tension of soft tissue through microincision in musculoskeletal disease.[39,40] Through these mechanisms, this treatment is expected to alleviate the spastic symptoms of CP. Besides, we previously reported case reports suggesting the clinical effect of acupotomy on post-stroke spasticity or cervical dystonia.[41,42] Because this novel treatment combines the effects of microsurgery and conventional acupuncture, based on the previously reported clinical effects of acupuncture on CP,[8–15] acupotomy may have a specific additive effect.

This study has the following limitations, and therefore, it is difficult to generalize the evidence derived. First, despite the comprehensive English, Chinese, and Korean database searches, only studies published in China were included, which may cause reporting bias and limit the generality of this review. Second, most included studies have low methodological quality. The information on the acupotomy procedure reported in the included studies was also insufficient. Third, although we planned subgroup analysis by the participants' age and follow-up period, we were unable to perform the analysis since all studies included children alone and only 1 study[29] conducted posttreatment follow-up. In addition, despite subgroup analysis according to primary data of TER and treatment period, heterogeneity still existed in some results. The wide age-range of the children included in each study and variability of the rehabilitation treatment as control intervention may have caused the heterogeneity. Fourth, in 7 studies,[28–34] conventional rehabilitation was concurrently used in both the experimental and control groups. Additionally, because of the invasive method of acupotomy, it is difficult to perform in blinded patients, which may lead to placebo effects and hence, the positive effects cannot be attributed to the efficacy of acupotomy alone. Furthermore, there was only 1 study[27] that compared the efficacy of acupotomy with rehabilitation therapy which was inadequate to explain the efficacy of acupotomy as monotherapy for CP. Finally, because there was no case where more than 10 studies were included in the meta-analysis, we could not evaluate the publication bias through funnel plots.

Future clinical trials evaluating the efficacy of acupotomy should be conducted with the following considerations in mind. First, the larger sample size based on calculation and high-quality trials are needed to obtain a high level of evidence. Moreover, it is necessary to use validated assessment tools that sensitively assess the effectiveness of interventions in CP. Second, the procedure of acupotomy should be reported in full compliance with the standardized reporting methods such as the Standards for Reporting Interventions in Controlled Trials of Acupuncture.[43] Third, since acupotomy is an invasive procedure, systematic monitoring of safety is needed, and all adverse reactions associated with acupotomy should be reported. Finally, because CP is one of the costliest chronic childhood conditions with increasing life expectancies, there is an increased financial burden.[44] Therefore, an evaluation of the cost-effectiveness of the acupotomy should be performed.

5. Conclusion

According to the current evidence, acupotomy as a monotherapy or as adjunctive therapy to rehabilitation treatment might have benefits for treating CP. However, due to the small number of included studies, the lack of appropriate sample size, poor methodological qualities, and low quality of evidence for main findings, the findings of this review should be interpreted with great caution. Larger and more rigorous high-quality RCTs should be performed in this area.

Author contributions

Chan-Young Kwon and Boram Lee performed the literature search, study selection, data extraction, and assessed the quality of methodological qualities using risk of bias tool. Chan-Young Kwon and Boram Lee described the manuscript, and Gyu Tae Chang and Sang-Hoon Yoon critically reviewed the manuscript. All authors participated in the analysis and interpretation of data and approved the final paper.

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