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 an 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, P = .02; 97% CI 1.11–1.34, f2 = .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, f2 = 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 positive effects 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.

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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

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 nonpro¬gressive 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.\textsuperscript{[13]} 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,\textsuperscript{[8]} oxidative stress,\textsuperscript{[9]} and neuronal apoptosis,\textsuperscript{[10]} and improve neurobehavioral ability in animal models of CP. Clinical studies have also shown that acupuncture improves motor function,\textsuperscript{[11]} alleviates adductor muscle tension,\textsuperscript{[12]} and promotes the development of intelligence\textsuperscript{[13,14]} in children with CP; therefore, it seems to be promising for the treatment of CP.\textsuperscript{[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).\textsuperscript{[16]} Recent studies have shown that acupotomy is clinically effective in the treatment of several musculoskeletal pain conditions.\textsuperscript{[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 origin of the spasticity are in brain, the induced spasticity of the local muscles and joints can cause a vicious circle, limiting the movements and consequently aggravating the disability.\textsuperscript{[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.](Image)
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 was 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 homogenous 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.
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.
| Author, yr | Sample size (G1:G2:G3) | Mean age (range) | Condition | G1 intervention | G2 intervention | G3 intervention | Duration | Frequency | Outcomes |
|------------|------------------------|------------------|------------|----------------|----------------|----------------|----------|----------|----------|
| Yan (2006) | 105 (35:34:36)         | NR (2–4 yr)      | Spastic cerebral palsy | Acupotomy | Bobath | Acupotomy plus Bobath | 1 mo |          | TER (by MAS score) at postintervention |
| Zhao (2013) | 90 (45:45)             | G1: 36.6 ± 7.2 mo (24–45 mo) | Spastic cerebral palsy | Acupotomy plus rehabilitation | Rehabilitation | -- | 3 mo | Acupotomy: 1 session/mo Rehabilitation: NR |
| Liang et al (2014) | 54 (27:27) | G1: 46.7 ± 8.1 mo (NR) | Spastic cerebral palsy | Acupotomy plus rehabilitation | Rehabilitation | -- | 6 mo | Acupotomy: 1 session/mo Rehabilitation: NR |
| Xue (2015) | 56 (28:28)             | G1: 22 ± 7 mo (12–36 mo) | Cerebral palsy | Acupotomy plus casts | Casts | -- | 2 wk | Acupotomy: single session Casts: keeping for 14 da |
| Chen (2016) | 30 (10:10:10)           | NR (0–12 yr)      | Spastic cerebral palsy | Acupuncture | Bobath | Acupotomy plus Bobath | 3 mo |          | 1) Flexion angle (active) of ankle, knee, and hip joint at postintervention | 2) MAS score (gastrocnemius) at 1 yr f/u 3) GMFM score at 1 yr f/u 4) Locomotion score (walking or wheelchair) in FIM at postintervention |
| Ye (2016) | 54 (30:24)             | NR (4–12 yr)      | Spastic cerebral palsy | Acupotomy plus Bobath | Bobath | -- | 3 mo | Acupotomy: 1 session/da Bobath: 30 min/1 session/da Acupotomy: 1 session/2 wk |
| Naerbuli (2017) | 101 (51:50) | G1: 8 ± 1 yr (1–16 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/da Bobath: 4 5min/1 session/da |
| Tan (2018) | 40 (20:20)             | G1: 6.9 ± 0.5 yr (1–15 yr) | Spastic cerebral palsy | Acupotomy plus rehabilitation | Rehabilitation | -- | 6 mo | Acupotomy: 1 session/mo Rehabilitation: NR |

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,[28,31] 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 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[24–31,33,34] 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; I² = 77%) and GMFM score (MD, 12.62; 95% CI, 11.75–13.49; I² = 54%), and significantly decreased muscle tone of gastrocnemius measured by AS or MAS (MD –0.97, 95% CI –1.07 to –0.88; I² = 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; I² = 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; I² = 75%) were significant. There were no significant differences in the results of both short-term (RR, 1.24; 95% CI, 0.97–1.58; I² = 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, –0.90, 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; I² = 0%). For
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 down-grading 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 | Anticipated absolute effects (95% CI) | Sample size | Risk with rehabilitation | Risk with acupotomy plus rehabilitation | Relative effect (95% CI) | Effects model | I² value | Z value | P value | Quality of evidence (GRADE) | Comments |
|----------|--------------------------------------|-------------|--------------------------|-----------------------------------------|-------------------------|-------------|--------|--------|--------|---------------------------|----------|
| TER      |                                      | RCTs 5      | 310 (702–1000)           | 870 (1000)                             | RR 1.24 (1.01–1.52)     | Random      | 77%    |        |        | Low                       | Risk of bias (−1) |
| Subgroup set 1 by MAS score |                                 | 2           | 124 (534–1000)           | 882 (1000)                             | RR 1.65 (0.87–6.07)     | Random      | 95%    |        |        | Very low                  | Risk of bias (−1) |
| By clinical symptoms |                                      | 1           | 56 (893–1000)            | 964 (1000)                             | RR 1.08 (0.93–1.25)     | Random      | NA     |        |        | Very low                  | Risk of bias (−1) |
| By clinical symptoms, MAS score |                                 | 1           | 90 (800–1000)            | 952 (1000)                             | RR 1.19 (1.02–1.49)     | Random      | NA     |        |        | Very low                  | Risk of bias (−1) |
| By clinical symptoms, MAS, GMFM score |                             | 1           | 40 (700–1000)            | 903 (1000)                             | RR 1.29 (0.93–1.77)     | Random      | NA     |        |        | Very low                  | Risk of bias (−1) |
| Subgroup set 2 Short-term (<6 mo) |                                      | 4           | 270 (702–1000)           | 871 (1000)                             | RR 1.24 (0.97–1.58)     | Random      | 83%    |        |        | Moderate                  | Risk of bias (−1) |
| Long-term (≥6 mo) |                                      | 1           | 40 (700–1000)            | 903 (1000)                             | RR 1.29 (0.93–1.77)     | Random      | NA     |        |        | Low                       | Risk of bias (−1) |
| AS or MAS score (gastrocnemius) Total |                                      | 3           | 114 –                    | MD 0.97 lower (0.07–0.88 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) |
| By MAS score |                                      | 2           | 60 –                    | MD 0.9 lower (1.09–0.71 lower)         | −                      | Fixed       | 3%     |        |        | Low                       | Risk of bias (−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) |
| Long-term (≥6 mo) |                                      | 2           | 94 –                    | MD 0.98 lower (1.08–0.89 lower)        | −                      | Fixed       | 0%     |        |        | Low                       | Risk of bias (−1) |
| GMFM score |                                      | 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) |
| Long-term (≥6 mo) |                                      | 2           | 94 –                    | MD 12.49 higher (11.52–13.46 higher)   | −                      | Fixed       | 75%    |        |        | Low                       | Risk of bias (−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[11,33,34] 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[11] 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 | I² 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.
| 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 acupuncture | 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 | Gastronomus | Loose texture (practitioner) | NR | 6 | 1 session/mo, 6 mo | Rehabilitation | NR |
| Xue (2015)[30] | Routine anesthesia | Achilles tendon | [1] Bouncing sensation (practitioner) | NR | 1 | 1 session/2 wk, 2 wk | Casts | NR |
| Chen (2016)[31] | Local anesthesia | 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 | NR | [2] Abnormal sensation (patients) [3] Radiation-like sensation (patients) | 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 | No. 4 (NR) | 12 (<5 yr old), 6 (<5 yr old) | 1 session/Wk (<5 yr old), 1 | Bobath | NR |
| Naerbuli (2017)[33] | NR | Achilles tendon, posterior tibial tendon, peroneal tendon | 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 | 1 session/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, and compliance is one of the important factors for the choice of treatment. Although acupuncture is considered to be safe, 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. 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. In addition, Novak et al. 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 evaluating gross motor skills and only 1 evaluated self-care ability. Furthermore, in the 3 studies 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. 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. Because this novel treatment combines the effects of microsurgery and conventional acupuncture, based on the previously reported clinical effects of acupuncture on CP, 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 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 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 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. 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. 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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References

[1] Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl 2007;109:8–14.

[2] Oskou M, Coutinho F, Dykeman J, et al. An update on the prevalence of cerebral palsy: a systematic review and meta-analysis. Dev Med Child Neurol 2013;55:509–19.

[3] Nelson KB. Can we prevent cerebral palsy? N Engl J Med 2003;349:1765–9.

[4] Surveillance of Cerebral Palsy in Europe.Surveillance of cerebral palsy surveys and registers. Surveillance of Cerebral Palsy in Europe (SCPE). Dev Med Child Neurol 2000;42:816–24.

[5] Christensen D, Van Naarden Braun K, Doernberg NS, et al. Prevalence of cerebral palsy, co-occurring autism spectrum disorders, and motor functioning – autism and developmental disabilities monitoring network, USA, 2008. Dev Med Child Neurol 2014;56:59–65.

[6] Zhang YQ, Liu JP, Wang J, et al. Traditional Chinese medicine for treatment of cerebral palsy in children: a systematic review of randomized clinical trials. J Altern Complement Med 2010;16:375–95.

[7] Novak I, Mcintyre S, Morgan C, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. Dev Med Child Neurol 2013;55:885–910.

[8] Qi YC, Xiao XJ, Duan RS, et al. Effect of acupuncture on inflammatory cytokines expression of spastic cerebral palsy rats. Asian Pac J Trop Med 2014;7:492–5.

[9] Li SH, Sun HT, Wang YM, et al. Therapeutic effect of acupuncture treatment on ischemic hypoxic neonate rats with cerebral palsy. Zhongguo Ying Yong Sheng Li Xue Za Zhi 2015;31:473–6.

[10] Zhao P, Li E. Study of acupuncture combined with rat nerve growth factor on neurorehabilitation ability of cerebral palsy infant rats and its brain tissue growth and metabolism associated proteins. Zhongguo Zhen Jiu 2018;38:631–6.

[11] Zhang J, Xu K, Ruan Y. Impacts on motor function in the children of cerebral palsy treated with acupuncture and acupoint embedding therapy, Zhongguo Zhen Jiu 2015;35:901–4.

[12] Jin B, Zhao Y, Li N. Impacts on adductor muscle tension in children of spasmocitic cerebral palsy treated with acupuncture and acupoint embedding therapy, Zhongguo Zhen Jiu 2015;35:217–20.

[13] Zhongguo Xi Yi Jie He Za Zhi 2013;33:924–9.

[14] Zhang Y, Lan R, Liu ZH. Impacts on tiope deformaty and intelligent development in spasmocytic cerebral palsy treated with acupuncture at naoqing xue (extra). Zhongguo Zhen Jiu 2015;33:95–9.

[15] Yang C, Hao Z, Zhang LL, et al. Efficacy and safety of acupuncture in children: an overview of systematic reviews. Pediatr Res 2015;78:112–9.

[16] Zhu HZ. Summarization of acupotomology. System Engin Sci (Chin) 2006;8:1–5.

[17] Zheng Y, Shi D, Wu X, et al. Ultrasound-guided miniscalpel-needle release versus dry needling for chronic neck pain: a randomized controlled trial. Evid Based Complement Alternat Med 2014;2014:235817.

[18] Ding Y, Wang Y, Shi X, et al. Effect of ultrasound-guided acucloty vs electro-acupuncture on knee osteoarthritis: a randomized controlled study. J Tradit Chin Med 2016;36:450–5.

[19] Kim HJ, Jeon JH, Kim YJ. Clinical effect of acupotomy combined with Korean medicine: a case series of a herniated intervertebral disc. J Acupunct Meridian Stud 2016;9:31–41.

[20] Ma C, Wu S, Li G, et al. Comparison of miniscalpel-needle release, acucloty, stretching, and exercise, and that of fentanyl in myofascial pain syndrome. Clin J Pain 2010;26:251–7.

[21] Li S, Shen T, Liang Y, et al. Miniscalpel-needle versus steroid injection for plantar fasciitis: a randomized controlled trial with a 12-month follow-up. Evid Based Complement Alternat Med 2014;2014:164714.

[22] Stocco A, Stecco C, Ragghianti P. Peripheral mechanisms contributing to spasticity and implications for treatment. Curr Phys Med Rehabil Rep 2014;2:121–7.

[23] The Cochrane Collaboration. Chapter 8: assessing risk of bias in included studies. In: Higgins J, Green S, eds, Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0, 2011. Available at: http://handbook-5-1.cochrane.org/. Accessed August 2018.

[24] Balshem H, Helfand M, Schunemann HJ, et al. GRADE Guideline: 3Rating the quality of evidence. J Clin Epidemiol 2011;64:401–6.

[25] Borenstein M, Hedges LV, Higgins J, et al. A basic introduction to fixed-effect and random-effects models for meta-analysis. Res Synth Methods 2010;1:97–111.

[26] Murad MH, Monton VM, Ioannidis JPA, Guyatt G, Rennie D, Meade MO, Cook DJ, et al. Fixed-effects and random-effects models. Users’ Guide to the Medical Literature: A Manual for Evidence-Based Clinical Practice. 3rd ed.New York, America: McGraw-Hill; 2015.

[27] Yan BC, Qiang F, Zhao XL. Acupotomy for the treatment of spastic cerebral palsy. Chin J Rehabil Theory Pract 2006;12:806.

[28] Zhao B, Sun XD, Man YG. Effects of acupotomy therapy on movement function of children with spastic cerebral palsy. Chin Man Rehabil Med 2013;4:73–7.

[29] Liang C, Sun J, Wang Q, et al. Clinical observation of acupotomyology combined with conventional rehabilitation training in the treatment of 27 cases pediatric spastic cerebral palsy lower limb motor dysfunction. J Pediatr Tradit Chin Med 2014;10:39–42.

[30] Xue WX, Chen CX, Zhang HY. Clinical effects of acupotomy combined with casting attachment on 28 cases of cerebral palsy with equinus deformity. Jiu J Tradit Chin Med 2015;47:56–7.

[31] Chen NP, Ma JL, Zhong Q, et al. Clinical observation on acupotomy combined with rehabilitation training in treatment of lower extremity joint deformity in pediatric spastic cerebral palsy. Chin J Rehabil Med 2016;31:60–3.

[32] Ye JR, Zhao JF. Clinical observation on acupotomy of spastic cerebral palsy. World Latest Med Inf 2016;16:225.

[33] Naerbul B, Fang LN, Yuan HL, et al. Clinical study on needle knife plus rehabilitation training for treatment of cerebral paralyis with spasthespondoa. Shanghai J Acu-Mox 2017;36:1329–32.

[34] Tan Y. Clinical effect of acupotomy combined with rehabilitative training in treating pediatric spastic cerebral palsy with lower limb joint deformity. J Med Theor Pract 2017;30:3010–2.

[35] Gaskell S. Evidence-Based Guidelines for the Management of Invasive and/or Distressing Procedures with Children. Leicester: The British Psychological Society; 2010.

[36] White A. The safety of acupotomology – evidence from the UK. Acupunct Med 2006;24(suppl):S33–7.

[37] World Health Organization.International Classification of Functioning, Disability and Health. Geneva: World Health Organization; 2001.

[38] Vargus-Adams JN, Martin LK. Measuring what matters in cerebral palsy: a breadth of important domains and outcome measures. Arch Phys Med Rehabil 2006;24(suppl):S53–7.

[39] Papavasiliou AS. Management of motor problems in cerebral palsy: a critical update for the clinician. Eur J Paediatr Neurol 2009;13:857–96.