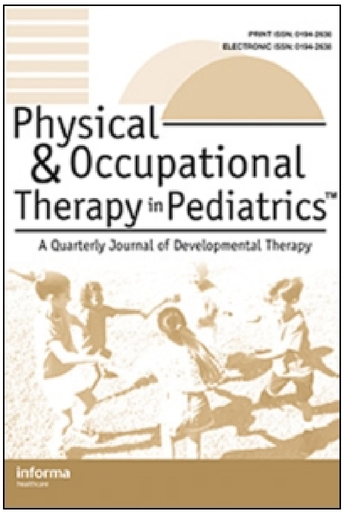

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Effects of Electrical Stimulation, Exercise Training and Motor Skills Training on Strength of Children with Meningomyelocele: A Systematic Review

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ABSTRACT. This systematic review provides a critical synthesis of research regarding the effects of electrical stimulation, exercise training, and motor skills training on muscle strength in children with meningomyelocele. Nine databases were searched using terms related to meningomyelocele and physical therapy interventions. Of 298

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potentially relevant citations, six met the inclusion criteria. Each was rated using the systematic review guidelines of the American Academy for Cerebral Palsy and Developmental Medicine. Two studies examined changes in quadriceps muscle torque following electrical stimulation, three investigated upper extremity exercise training, and one evaluated quadriceps strength after motor skills training. Although the limited evidence suggests improvements in strength when using these interventions, much of the evidence is of low methodological quality and all studies were published more than 10 years ago. Further research is needed regarding various strength-training interventions for children with meningomyelocele and the relationship between increased strength and improved activity and participation.

KEYWORDS. Meningomyelocele, spina bifida, exercise training, motor skills training, electrical stimulation, muscle strength

The term *spina bifida* incorporates a range of congenital abnormalities that involve incomplete closure of the spinal column (Hinderer, Hinderer, & Shurtleff, 2006); spina bifida occurs in approximately 0.5–1.0 in 1000 pregnancies annually in the United States, despite the fact that use of folic acid before conception has decreased the occurrence of open neural tube defects (Shaer, Chescheir, & Schulkin, 2007). Of these individuals, 94% have meningomyelocele (MM), a form of spina bifida that involves a protrusion of the spinal cord through an opening of the vertebral column and typically results in some paralysis in the lower extremities.The resulting impairments depend on the level of the lesion and can include muscle weakness, paralysis, loss of lower extremity sensation, loss of bladder or bowel function, and learning disabilities (Hinderer et al., 2006). Fortunately, intrauterine surgery on infants identified with MM (via prenatal ultrasound) can reduce the need for shunting for hydrocephalus and the incidence of urinary tract infections and can preserve lower extremity function present at the time of the repair (Bruner, 2007).

Children with MM typically receive a multidisciplinary treatment approach. Physical therapists assist with managing the multiple systems affected and preventing further complications. Typical physical therapy (PT) interventions center around a goal of optimizing mobility and maximizing independence that can be facilitated by muscle strengthening, adaptive positioning, and/or enhancing postural control (Hinderer et al., 2006; Ryan, Ploski, & Emans, 1991).

We determined that examining the effects of muscle strengthening interventions for children with MM was important because prior studies have shown that strength (Agre et al., 1987) or endurance training (O'Connell, Barnhart, & Parks, 1992) can enhance functional outcomes in these children (O'Connell & Barnhart, 1995). Interventions aimed at muscle strengthening from the research literature have included electrical stimulation (Karmel-Ross, Cooperman, & Van Doren, 1992; Mazliah, Naumann, White, Milner, & Carroll, 1983), behavioral treatments (Manella & Varni, 1984; Rapport & Bailey, 1985), and motor skills training (Manella & Varni, 1984).

Evidence regarding the efficacy of these PT interventions in children with MM is limited. After a comprehensive search of the literature, only one systematic review examining lower limb bracing and ambulation training for children with thoracic or upper lumbar level lesions (Mazur & Kyle, 2004) was located. The authors of this review concluded that controversy existed within the literature regarding the use of ambulation with bracing versus wheelchair use for children with MM and that more research was necessary.

In contrast to the limited research literature on PT interventions for children with MM, there is a much larger body of research regarding cerebral palsy, another common motor disorder in children. In a 2002 systematic review on the effectiveness of strength training for children with cerebral palsy, 23 relevant articles were included, with 11 of high methodological quality (Dodd, Taylor, & Damiano, 2002). This gap in literature is of great importance as physical therapy plays a major role in promoting activity and participation for children with MM (Hinderer et al., 2006) and there is little evidence to guide best practice.

One purpose of a systematic review is to "gather and present the best evidence for or against the effectiveness of an intervention" (O'Donnell et al., 2004). Reviews can also help identify gaps in the literature and new areas of research that are needed (O'Donnell et al., 2004). In particular, the purpose of this review is to provide a critical synthesis of the available research that investigated the effects of strengthening or resistance interventions on muscle strength in children with MM.

LITERATURE SEARCH

The search strategy for this review included the following databases: PEDro, CCRT, DARE, CIRRIE, Cochrane Database of Systematic Reviews, PubMed, EMBASE, CINAHL, and MEDLINE, searching from the inception of each database through December 2007. Articles retrieved were limited to studies published in the English language. *Meningomyelocele* and *physical therapy* were the main search terms and relevant terms and synonyms were also explored, i.e., myelodysplasia, myelomeningocele, spina bifida, exercise, rehabilitation, strength training, resistance training. The reference lists of selected articles were searched for potentially relevant citations. The first five authors, all students in an entry-level master's in physical therapy program, participated in the literature search with each database being explored by one author.

Inclusion and Exclusion Criteria

Articles were initially screened by title, then by abstract, and finally by the full text. Studies had to fulfill the following criteria to be included in this review: (1) participants diagnosed with MM; (2) participants 21 years of age or under; and (3) the study involved a PT intervention. Book chapters, duplicate publications, or studies that focused on the following variables were excluded: surgical techniques, orthoses and bracing, bowel and bladder function, cognition or scoliosis. The first five authors took part in this process with two examining each article for its potential relevance. If reviewers were uncertain of the article's relevance, it was kept for the next screening process. Disagreements regarding inclusion or exclusion of an article were settled by a third reviewer.

Due to the limited number of articles acquired through the search process, inclusion and exclusion criteria were kept broad. However, after data extraction and initial analysis of the full text, the dependent variables for the 11 initially included studies were extremely heterogeneous, making data synthesis impractical. Outcomes in the initial 11 studies ranged from strength and cardiopulmonary function to functional mobility. For the purposes of this review, the inclusion criteria were further narrowed to require strength, defined as "the maximum force or tension generated by a single muscle or related muscle groups" (McArdle, Katch, & Katch, 2001, p. 501), as an outcome, thus allowing for a more homogeneous group of studies. The participants were all children, 21 years of age and under, with MM. The independent variables considered in this review were electrical stimulation, exercise training or motor skills training, with strength of relevant muscle groups as the common dependent variable or outcome.

Quality Assessment

Studies selected for this review were assigned levels of evidence based on the AACPDM Levels of Evidence (O'Donnell et al., 2004; Table 1) and

TABLE 1. American Academy for Cerebral Palsy and Developmental Medicine Levels of Evidence (O'Donnell et al., 2004)

Level	Intervention (Group) Studies
I	Systematic review of randomized controlled trials (RCTs)
	Large RCT (with narrow confidence intervals) ($n > 100$)
II	Smaller RCTs (with wider confidence intervals) ($n < 100$)
	Systematic reviews of cohort studies
	"Outcomes research" (very large ecologic studies)
III	Cohort studies (must have concurrent control group)
	Systematic reviews of case control studies
IV	Case series
	Cohort study without concurrent control group (e.g., with historical control group) Case-control study
V	Expert opinion
	Case study or report
	Bench research
	Expert opinion based on theory or physiologic research
	Common sense/anecdotes

were categorized into one of the following three levels: II, IV, or V. The AACPDM quality assessment, presented in Table 2, was used to evaluate the quality of the one article included in the level II group (O'Donnell et al., 2004). Because five of the six included studies were either case series or case reports and thus are not recommended for quality assessment using the AACPDM criteria, their rigor was assessed using a set of quality questions presented in Table 3 that were developed for case studies by authors of a 2006 (unpublished) systematic review (Dean et al., 2006). Studies were determined to be of strong quality if they obtained a score of 6 or 7, of moderate quality with a score of 4 or 5, and of weak quality with a score of 3 or less (O'Donnell et al., 2004). The quality assessment scores for the included studies are presented in Table 4. These ratings were completed for each article by two independent reviewers. The level of agreement for assigning levels of evidence was 100% and the level of agreement for determining quality of studies was 83%.

Data Extraction and Analysis

A standardized data extraction form was developed and used to collect relevant information from each article, i.e., author and year; database;

TABLE 2. American Academy for Cerebral Palsy and Developmental Medicine: Assessing Quality of Conduct of a Study (O'Donnell et al., 2004)

Questions:
1. Were inclusion and exclusion criteria of the study population well described and followed?
2. Was the intervention well described and was there adherence to the intervention assignment? For 2-group designs, was the control exposure also well described?
3. Were the measures used clearly described, valid, and reliable for measuring the outcomes of interest?
4. Was the outcome assessor unaware of the intervention status of the participants (i.e., were there blind assessments)?
5. Did the authors conduct and report appropriate statistical evaluation including power calculations?
6. Were dropouts/loss to follow-up reported and less than 20%? For 2-group designs, was dropout balanced?
7. Considering the potential within the study design, were appropriate methods for controlling confounding variables and limiting potential biases used?

number of groups in the study; age, gender, and level of impairment of participants; type, frequency, and duration of intervention; outcome measures; adherence/dropout; and any adverse effects. Data extraction was completed independently by two reviewers. If there was any disagreement on the data extracted, the two reviewers discussed this until resolution

TABLE 3. Case Study Methodological Quality Assessment Tool (Dean et al., 2006)

Questions:
1. Was the purpose of the study clearly stated?
2. Was the hypothesis clearly stated?
3. Were the patients described in detail so that you could decide whether they are comparable to those seen in practice?
4. Were the interventions and treatment settings described well enough so that they could be replicated?
5. Were the measures used clearly described, valid, and reliable for measuring the outcome of interest?
6. Was the size of the effect clinically important?
7. Were the limitations of the study identified and discussed?

TABLE 4. Study Quality Assessment

Study	Level	1	2	3	4	5	6	7	Total[b]
[a]AACPDM: Assessing Quality of Conduct of a Study (O'Donnell et al., 2004)									
Andrade et al. (1991)	II	√	√	√				√	4
Quality Assessment of Case Reports (Dean et al., 2006)									
Karmel-Ross et al. (1992)	IV	√		√	√	√	√	√	6
Mazliah et al. (1983)	IV	√	√	√	√			√	5
O'Connell & Barnhart (1995)	IV	√		√	√		√	√	5
Manella & Varni (1984)	V	√		√	√	√	√		5
Rapport & Bailey (1985)	V	√		√	√	√	√	√	6

[a]AACPDM = American Academy for Cerebral Palsy & Developmental Medicine.
[b]Strong ("yes" score on 7 or 6), moderate (score 5 or 4), or weak (score $\leq$3).

was achieved. Relevant information was entered into summary tables for further investigation (Tables 5 and 6).

Data analysis was conducted using a descriptive synthesis method (Centre for Reviews and Dissemination, 2001). This method includes evaluating the participants, interventions, and outcomes of the studies to determine whether findings can be generalized for the greater population. Limitations are investigated and missing components are noted in order to progress the understanding of the present research base and facilitate the direction of future research. A meta-analysis was not conducted due to the variability of research designs and the heterogeneity of outcome measures and ages of participants.

Summary of Studies

The literature search resulted in 298 potentially relevant citations. After screening the titles, 138 articles remained that appeared to fulfill inclusion and exclusion criteria. Based on abstract screening, 120 articles were discarded because they, in fact, failed to satisfy the inclusion/exclusion criteria, leaving 18 for full text review. Hand searching resulted in two more articles being found eligible for full text retrieval. After full text review of the 20 relevant articles, 11 satisfied the initial inclusion and exclusion criteria. After modification of screening criteria to enhance study homogeneity, five studies were excluded that did not include strength as an outcome measure. This process of study selection resulted in six articles to be included in this systematic review (Figure 1).

TABLE 5. Summary of Study Interventions

Study citation and description	Therapy intervention	Control intervention	Sample
Andrade et al. (1991) Pretest-posttest with nonrandomized control group Level II Quality 4/7	Exercise program Aerobic and UE strengthening components 1 hr/week for 10 weeks Isotonic or dynamic exercises involving the shoulder flexor and abductor muscles, elbow flexors and extensors, and abdominal muscles	Children not attending exercise program	*N* = 13 (7 males; 6 females) Experimental group (*N* = 8); control group (*N* = 5) Children with MM; lesion below T6, 8–13 years
Karmel-Ross et al. (1992) Pretest-posttest control group design (case series) Level IV Quality 6/7	Electrical stimulation Applied to quadriceps femoris of randomly assigned limb in conjunction with walking/standing activities 30-min sessions, 6x/week for 8 weeks. Maximum current of 50 mA delivered at 35 pulses per second	Contralateral limb	*N* = 5 (2 males; 3 females) Children with SB; lesion at level L2-3, 5–21 years
Manella & Varni (1984) Case report Level V Quality 5/7	Motor skills training & behavioral therapy Functional activities (standing and gait) Clinical program: 30 min sessions for 4 weeks; home program: 30 min daily; follow-up: 1x/month for 5 months	Not applicable	*N* = 1 (female) Child with MM at L3, 5 years
Mazliah et al. (1983) One-group pretest-posttest design (case series) Level IV Quality 5/7	Electrical stimulation Applied to quadriceps femoris bilaterally 1-2 hr sessions daily for 6 months. Stimulus trains of 15 s on and 25 s off at a frequency of 50 Hz	Not applicable	N = 3 Children with lumbar MM Knee flexion contractures $>15^\circ$, 9–12 years

(Continued on next page)

TABLE 5. Summary of Study Interventions *(Continued)*

Study citation and description	Therapy intervention	Control intervention	Sample
O'Connell & Barnhart (1995) One group pretest-posttest design (case series) Level IV Quality 5/7	Exercise program UE strengthening 30 min sessions 3x/week for 9 weeks. Training involved three sets of 6-RM exercises for elbow flexors and extensors; shoulder flexors, extensors, abductors, internal, and external rotators; and a combined shoulder flexion-elbow extension exercise performed in supine	Not applicable	$N = 6$ Children diagnosed with MM (N = 3), lesion at T12 ($N = 2$), and T8 ($N = 1$) Children diagnosed with CP ($N = 3$), 4–16 years
Rapport & Bailey (1985) SSRD (multiple baseline across behaviors) Level V Quality 6/7	Exercise program & behavior therapy Fine and gross motor activities and use of positive reinforcement Clinical program: 90-min sessions 1x/week for 6 weeks; home program: 30 min/day, 5x/week for 56 weeks	Not applicable	$N = 1$ (male) Child with MM, 8.5 years

Abbreviations: Cerebral palsy = CP, lumbar = L, meningomyelocele = MM, single-subject research design = SSRD, spina bifida = SB, thoracic = T, upper extremity = UE.

Table 5 provides a summary of the six articles included in this systematic review. The levels of evidence of the included studies were II, IV, or V and the quality of studies ranged from 4 to 6. Of the six studies that investigated the effects of therapy interventions on strength, two examined electrical stimulation (Karmel-Ross et al., 1992; Mazliah et al., 1983), three used exercise programs (Andrade, Kramer, Garber, & Longmuir, 1991; O'Connell & Barnhart, 1995; Rapport & Bailey, 1985), and one assessed motor skills training (Manella & Varni, 1984).

TABLE 6. Summary of Study Results

Study	Outcome of Interest	Measure	Result
Andrade et al. (1991)	Self-concept	Harter Self-Perception Profile for Children	
	Cardiovascular endurance	Modified AAHPERD 9-min run (Winnick & Short 1984b)/ Heart rate	$p < .01$ NS
	Strength	Isometric tests using handheld dynamometer	$p < .01$
Karmel-Ross et al. (1992)	Quadriceps muscle strength	Maximum isometric voluntary knee extension torque	NS for 3 subjects; $p < .05$ for 2 subjects
	Functional tasks (timed)	80 ft walking & 20 steps (ascending and descending)	4 subjects improved; 1 subject did not improve
Manella & Varni (1984)	Achieving independence in motor skills through behavioral physical therapy	Distance walked	Increased from 10 ft to 90 ft within the trial (to 400 ft at follow-up).
		Number of steps	Increased from 5 to 100.
		Quadriceps manual muscle test	Increased from Fair to Good.
Mazliah et al. (1983)	Quadriceps muscle strength	Isometric torque measurements	Improved ($n = 2$)
	Quadriceps girth	Measuring tape	NS
	Knee flexion contracture	Goniometry	Improved ($n = 1$)
	Gait pattern	Dynamic gait assessment	NS
O'Connell & Barnhart (1995)	Wheelchair propulsion	50-m wheelchair propulsion test	NS
		12-min wheelchair propulsion test	$p = .031$
		6-repetition maximum	$p = .018$ to $.031$
Rapport & Bailey (1985)	Arm/hand fine motor function	Southern California Motor Accuracy Test	Gradual improvement based on graphed data

(Continued on next page)

TABLE 6. Summary of Study Results *(Continued)*

Study	Outcome of Interest	Measure	Result
	Arm/hand gross motor function	Osco pinch meter	Gradual improvement based on graphed data
		Minnesota Rate of Manipulation Test	Rapid improvement based on graphed data
		Dynamometer grip strength	Accelerated improvement based on graphed data

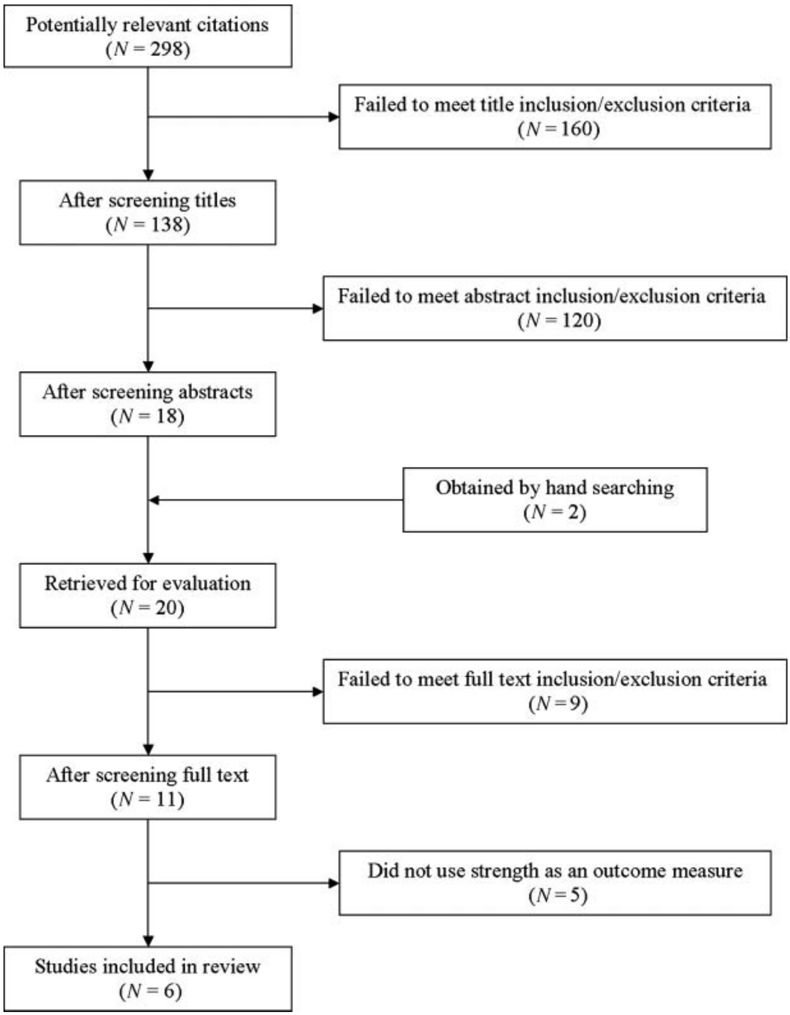
Note. NS = Not significant, AAHPERD = American Alliance for Health, Physical Education, Recreation and Dance.

The two studies that used electrical stimulation as an intervention included a one-group pretest-posttest study (or case series) by Karmel-Ross et al. (1992) with level IV evidence of strong quality. The second electrical stimulation study by Mazliah et al. (1983) used a one-group pretest-posttest design (case series) and provided level IV evidence of strong quality. In both studies, the outcome assessed was lower extremity strength. The three studies that investigated the effects of exercise programs on upper extremity strength included a pretest-posttest control group design by Andrade et al. (1991), a study providing level II evidence of moderate quality, a one-group pretest-posttest design (case series) by O'Connell and Barnhart (1995) providing level IV evidence of moderate quality, and a 1985 single-subject multiple baseline design across behaviors by Rapport and Bailey (level V evidence of moderate quality). Manella and Varni(1984) also implemented a single-subject multiple baseline design across behaviors (level V evidence of strong quality) to explore the effects of motor skills training in conjunction with behavioral therapy on strength.

SUMMARY OF RESULTS

The investigated outcomes, measures used, and results of the articles included in this systematic review are summarized in Table 6. The total number of individuals with MM across all six studies was only 26, with the largest study including 13 participants (Andrade et al., 1991). Overall, the children's ages ranged from 4 to 21 years and lesion levels were all below T6.

FIGURE 1. Screening process.



Electrical Stimulation

Karmel-Ross et al. (1992) and Mazliah et al. (1983) investigated the effects of electrical stimulation on quadriceps muscle strength. Isometric torque measurements of knee extension were used to determine the effectiveness of interventions but the protocols of these two studies varied. The five participants in the Karmel-Ross et al. study ranged in age from 5 to 21 with lesion levels at L2-3. Thirty minutes of electrical stimulation was provided six times per week for eight weeks to one of the lower extremities of each participant. One electrode was placed over the belly of the vastus lateralis and rectus femoris muscles and another over the rectus femoris and vastul medialis. A portable neuromuscular stimulator "was programmed to produce rectangular, biphasic, symmetrical pulses with durations of 347 microseconds per phase; a maximum current of 50 mA delivered at 35 pulses per second; and a 2-second up and 5-second down ramp" (p. 724). During week 1 of the study, the stimulator was on for 8 s and then off for 24 s. During the second week, the stimulator was on for 8 s and off for 16 s, and during weeks 3 to 8, the on/off times were 8 s each. With the contralateral limb serving as the "control," Karmel-Ross and colleagues found statistically significant improvements of knee extension torque in two of five participants. No adverse effects were experienced by any of the five participants. In relating their results to potential functional (participation) outcomes, the authors concluded that electrical stimulation may be effective "for increasing the ambulatory potential" for some children with MM.

In the Mazliah et al. (1983) study, there were three children, ages 9 to 12, all with lumbar-level lesions. In contrast to Karmel-Ross et al. (1992), Mazliah and colleagues used electrical stimulation bilaterally with no control condition. The stimulation was applied 1 to 2 hr daily for six months using stimulus trains of 15 s on and 25 s off at a frequency of 50 Hz. Two of the three participants demonstrated improvements in knee extension torque.

In the study by Karmel-Ross et al. (1992), health issues and a lack of patient adherence were stated to contribute to the low number of significant findings.The study by Mazliah et al. (1983) required the family or participant to implement the electrical stimulation, which may have increased the risk of low adherence or ineffective application. These two studies suggest that quadriceps torque can be increased by electrical stimulation in some children with MM, which could potentially lead to enhancing these children's participation by improving their ability to ambulate.

Exercise Training

Andrade et al. (1991) and O'Connell and Barnhart (1995) reported significant improvements in upper extremity strength after resistance training. Andrade et al.'s level II study included 13 children, aged 8 to 13 years, with lesions below T6. These authors implemented a once-weekly, 10-week program, which included upper extremity progressive resistance training in combination with warm-up, aerobic training, and cool down for 1 hr per week. The strength training employed isotonic or dynamic exercises involving the shoulder flexor and abductor muscles, the elbow flexors and extensors, and the abdominal muscles; exercises were performed for one set of 10 repetitions. Using a hand-held dynamometer to measure strength, statistically significant results were obtained by the experimental group compared to the control group.

Participants in the O'Connell and Barnhart (1995) study were children with MM ($n = 3$) or CP ($n = 3$) ranging in age from 4 to 16 years. One child with MM had a T8 lesion and the other two had lesions at T12. This study was of similar duration (nine weeks) to that by Andrade and colleagues, but included only resistance training in 30-min sessions three times per week. The training involved three sets of six-repetition maximum (6-RM) exercises for elbow flexors and extensors; shoulder flexors, extensors, abductors, and internal and external rotators; and a combined shoulder flexion-elbow extension exercise performed in supine. Statistically significant results were attained using 6-RM testing. These two studies' results suggest that resistance-training programs can provide an adequate stimulus to increase upper extremity strength in children with MM. Other important outcomes included enhanced cardiovascular endurance and self-concept (Andrade et al., 1991) and significantly increased distance covered in a 12-min, wheelchair, distance test (O'Connell & Barnhart, 1995).

In a single-subject design, Rapport and Bailey (1985) investigated the effects of a 1 hr per week, clinic-based PT program for six weeks that was then combined with a 56-week home program that focused on fine motor coordination and hand strength, e.g., game-type tasks and hand-grip activities. The home program was carried out for 30-min sessions, five days a week. Improvements in grip and pinch strength were demonstrated using a hand held dynamometer and a pinchmeter, with the greatest improvements noted during the combined clinic-based and home program. The motivational component to this program, i.e., a daily chart with rewards for the child's efforts, as well as the involvement and cooperation of the parents, were considered key elements with regard to the positive therapeutic outcomes.

Motor Skills Training

Based on manual muscle testing, Manella and Varni (1984) reported improvement in quadriceps strength from grade 3 (Fair) to a grade 4

(Good) bilaterally after four weeks of motor skills training combined with behavior therapy in a 5-year-old girl with MM at L3. The motor skills practice included sit to stand, standing erect, ambulation in parallel bars, and three-point gait using a rolling walker. There were 19 daily 30-min treatment sessions over a four-week period. Behavior therapy included social and food reinforcers to encourage participation in the functional-based activities, which were gradually removed over the four-week period. This single-subject design suggested effectiveness of motor skills training as well as the value of behavior therapy in pediatric health care delivery. The fact that this child's distance ambulating at home increased from 15 feet midway through the course of the study to 400 feet during the post-study, follow-up period suggests that the motor skills practice resulted not only in increased strength but, more importantly, functional household ambulation.

DISCUSSION

Based on this systematic review, the evidence suggests the possibility of being able to increase muscle strength using electrical stimulation, exercise training, or motor skills training for children with MM. All six studies or case reports showed improvements in strength with three reporting statistically significant improvements; there were no adverse effects noted. Caution needs to be taken when interpreting the results of this limited number of findings, especially when lacking results from more rigorous types of studies. Recommendations directing therapists to optimal treatment protocols are not possible due to the small number and variability across the studies. Recommendations could certainly change or become clearer when stronger evidence is available.

Several limitations are recognized within this systematic review. The search strategy was limited to English and published articles resulting in only six studies of lower evidence levels. Gray literature (e.g., technical reports, theses, government documents) was not extensively searched, possibly overlooking current information. Data synthesis was completed using a descriptive synthesis method versus a meta-analysis due to the variability of research designs, intervention protocols, ages of participants, and outcome measures used. All of the studies were limited by small sample sizes. The studies were not published recently, the most recent being completed in 1995 (O'Connell & Barnhart). And finally, the criteria used to assess the study quality of case reports (Dean et al., 2006) have not been validated.

Credible results arise from studies of high levels of evidence and quality, which enables generalizability and applicability of treatment approaches to the population of interest. This systematic review included only one study of level II evidence, investigating the effects of an exercise program on strength (Andrade et al., 1991). The remaining five studies were of only level IV or V evidence. Therefore, results of the studies included in this systematic review should be applied carefully to the general population of children with meningomyelocele.

Despite these limitations, these studies reveal important aspects related to interventions focused on strengthening for children with MM and provide valuable information. While the levels of evidence are generally lower, our quality assessment revealed that the studies were of strong (Karmel-Ross et al., 1992; Rapport & Bailey, 1985) or moderate quality (Manella & Varni, 1984; Mazliah et al., 1983; O'Connell & Barnhart, 1995). Valuable components of the available research include the investigation of both lower and upper extremity muscles using various methods (e.g., electrical stimulation for lower extremity muscles and strength training for upper extremity) and the value of relating strength gains to functional performance. Strengths of this systematic review include an extensive literature search, which was supplemented with monthly updates, the use of five reviewers to limit biases during data extraction and analysis, access to an expert in the field, and excellent inter-rater percentage agreement.

Areas for future research include further investigation of the potential effects of electrical stimulation on ambulation ability, as introduced in the study by Karmel-Ross et al. (1992), as well as the prevention of knee flexion contractures, discussed by Mazliah et al. (1983). In particular, the appropriate dosage of electrical stimulation requires further study, as it appears that the higher dose used by Karmel-Ross et al. may have been more effective. Although literature regarding strength training for children with MM is limited and was published before 1996, an abundance of recent studies have examined different strengthening interventions for children or adolescents with spinal cord injury (e.g., Johnston, Betz, Smith, & Mulcahey, 2003; Johnston et al., 2003; Johnston et al., 2005; Johnston, Smith, Oladeji, Betz, & Lauer, 2008) and cerebral palsy (e.g., Katz, Tirosh, Marmur, & Mizrahi, 2008; Morton, Brownlee, & McFadyen, 2005; Stackhouse et al., 2007; Verschuren, Ketelaar, Takken, Helders, & Gorter, 2008). Evaluation of some of these interventions, such as implanted functional electrical stimulation, is certainly warranted for children with MM.

It may be beneficial for physical therapy researchers to investigate additional areas in the treatment approach such as adherence, motivation, effective education, and the importance of parental involvement during treatment when applying strength training principles to children with MM. Rapport and Bailey (1985) discussed the importance of program duration and emphasized the value of allowing time for practice and perseverance, especially in children with disabilities. Most importantly, future studies should examine the effects of strengthening on activity and participation in children with MM.

Children with MM interact with a multitude of healthcare professionals, including physical therapists. Although this review supports aspects of physical therapy interventions, the literature reviewed did not allow for conclusive recommendations. In order to ensure physical therapy is delivered in an effective and efficient manner, further, more rigorous research is necessary.

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