Self-concept in children with spina bifida compared with typically developing children

Review

Nora Shields* PhD;
Nicholas F Taylor PhD;
Karen J Dodd PhD, Musculoskeletal Research Centre and the School of Physiotherapy, La Trobe University, Melbourne, Victoria 3086, Australia.

**Correspondence to first author at* School of Physiotherapy, Division of Allied Health, La Trobe University, Victoria 3086, Australia.
E-mail: N.Shields@latrobe.edu.au

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The literature was systematically reviewed to determine if children with spina bifida have lower self-concept compared with their peers with typical development. Relevant trials were identified by searching electronic databases, supplemented by citation tracking. Of 803 papers initially identified, 15 met the inclusion criteria. Meta-analysis revealed children with spina bifida scored significantly lower than children with typical development for the domains of global self-worth (*d*=−0.39, 95% confidence interval [CI] −0.65 to −0.12); physical appearance (*d*=−0.26, 95% CI −0.46 to −0.06); athletic competence (*d*=−0.45, 95% CI −0.67 to −0.22); social acceptance (*d*=−0.33, 95% CI −0.55 to −0.11); and scholastic competence (*d*=−0.43, 95% CI −0.66 to −0.21). There was no difference between the groups for the behavioural conduct domain. Children with spina bifida on average have a lower self-concept than their peers with typical development. Clinicians need to take account of this information in planning the assessment and treatment of this group.

Self-concept is a multidimensional psychological construct that describes what children think of themselves in domains such as social acceptance, athletic competence, scholastic competence, behavioural conduct, and physical appearance. It comprises the child's perceived identity or their awareness of their own personal characteristics and attributes, and their global sense of self-worth (self-esteem) or how they evaluate their characteristics in relation to others.[1] Self-concept is a fundamental component of a child's psychological health and development.[2]

Spina bifida is a congenital neural tube defect affecting 1 in every 1000 live births.[3] The degree of impairment varies and is dependent on the level and extent of the lesion and the amount of neural tissue involved. Typical impairments include muscle weakness or paralysis, sensory deficits, cognitive deficits, musculoskeletal deformities, and urinary and bowel incontinence,[4] all of which may cause difficulties in everyday functional activities such as walking, dressing, and other tasks of personal care. These impairments could conceivably have an impact on the self-concept of children with spina bifida,[5] by limiting the child's ability to explore and interact with others and their environment.[6,7] However, there is conflicting evidence on whether children with spina bifida indeed have a lower self-concept.[2,5,6,8]

Health professionals need to consider the child's views when deciding optimal management strategies, selecting interventions, and planning for the future.[9,10] Understanding the child's self-concept is an important component of taking account of their views. Recognition of the self-concept of children with spina bifida may also contribute to and enhance the development of the child–clinician relationship. It may help clinicians identify children at risk of lower self-concept,

thus facilitating appropriate referral or providing anticipatory guidance.[6]

To assist clinicians in this regard, a systematic review was conducted to find out if the self-concept of children with spina bifida was different from that of children with typical development. The aims of the review were: (1) to compare the self-concept of children with spina bifida with that of children with typical development for the domains of: global self-worth, physical appearance, athletic competence, social acceptance, behavioural conduct, and scholastic competence; and (2) to investigate if there are differences in self-concept between age, sex, and socioeconomic status.

Method

SEARCH STRATEGY

Relevant literature was sourced by searching the following electronic databases: AMED (1985–April 2008), CINAHL (1982–April 2008), EMBASE (1988–April 2008), ERIC (1966–April 2008), Medline (1966–April 2008), PEDro (1929–April 2008), PsycINFO (1872–April 2008), PubMed (1966–April 2008) and the Cochrane library. The keywords used were: 'spina bifida' or 'myelomeningocele' combined with 'child' and 'self-concept'. Relevant synonyms for each term were incorporated into the search strategy (e.g. self-esteem, self-efficacy, and self-perception were used for self-concept). Reference lists of the identified articles were manually searched to identify additional relevant articles. Citation tracking of the included studies and key authors in the area was also conducted using the Web of Science.

INCLUSION/EXCLUSION CRITERIA

The following criteria were applied by two independent assessors (NS, YL) to the titles and abstracts of the search yields. Studies were selected if they included children with spina bifida aged under 18 years, compared the self-concept of children with spina bifida with typically developing peers, and measured self-concept using a quantified scale. Studies were excluded if any of the participants had a condition other than spina bifida (e.g. cerebral palsy), focused on outcomes that measured a construct other than self-concept (such as, functional assessment or quality of life), or collected data by qualitative methods (such as unstructured or semi-structured interviews). There was no language restriction. In cases where the title or abstract provided insufficient information about the study, the full text of the article was obtained and read by both assessors. Discrepancies in the decisions made were discussed until a consensus was reached.

QUALITY ASSESSMENT

The quality of the studies selected for review was rated using five criteria specific to assessment of observational studies (Table I) adapted from Khan et al.[11] These criteria were chosen to ensure the included studies demonstrated an appropriate degree of internal validity and provided the relevant statistics to allow accurate conclusions to be drawn. Each item was scored as either met (two points), partially met (one point) or not met (no points). All articles were assessed independently by two assessors (NT, KD). Both assessors were blinded to the source of the article including the authors and their affiliations, journal name, and publication date. Any disagreements between the two reviewers were resolved by discussion until a consensus was reached.

DATA EXTRACTION AND STATISTICAL ANALYSIS

Data were extracted from the included studies by two independent reviewers (NT, KD) using a standardized data extrac-

Table I: Quality assessment measure of selected studies

No	*Criterion*	*Satisfied if:*
1	Was the study based on a representative sample selected from a relevant population?	The report described the source and demographic details (sex, age, level of disability) of the participants with spina bifida and the source for the children with typical development. This item was partially met if only described in detail for children with spina bifida or only described some of the demographics of the children.
2	Were the criteria for inclusion and exclusion explicit for both the children with spina bifida and the children with typical development?	The report described a list of criteria to determine eligibility for the study for both children with spina bifida and children with typical development. This item was partially met if it only described in detail the criteria for the children with spina bifida.
3	Were the two groups comparable on all potential confounding factors?	The groups had to be directly comparable for sex and age. The reviewer had to also take into account if socioeconomic status and schooling were comparable. This item was partially met if age and sex were comparable but other factors might have been confounding.
4	Did the outcome measurement tools used demonstrate sufficient validity for comparing the self-concept of the groups?	This item was met if the self-concept measurement tool was referenced and either stated that the tool has demonstrated evidence of validity with a reference, or reports that the tool had been able to detect changes or differences in a relevant population (e.g. children with physical disabilities) with a reference.
5	Was an appropriate statistical analysis used?	This item was partially met if only means and SDs were reported. This item was partially met if it is just reported that the tool has good measurement properties or reliability, without mention of validity. The report provided means and SDs (or medians and interquartile range) for each group for the measure of self-concept, and reported an appropriate statistical test for comparison (*t*-test or equivalent non-parametric test such as Mann-Whitney *U*).

tion form developed for the review. Details of the study objective, study design, participant characteristics (including age, sex, and background), outcome measures used, main results, and study limitations were documented.

Standardized mean differences (effect sizes *d*) were calculated by subtracting the mean of the children with typical development from the mean of the group with spina bifida, and dividing by the pooled standard deviation (SD). Accordingly, the included studies needed to report the means and SDs for both groups of children. Calculations were completed using web-based software.[12] Standardized mean differences less than 0.20 were considered small, between 0.20 and 0.50 were considered medium, and greater than 0.80 were considered large.[13]

Meta-analysis was conducted to provide an overall estimate of the effect for each of the domains where outcomes were considered similar enough to pool. To account for any heterogeneity in the data, a random effects model was applied, as this model assumes variability due to a combination of random sampling error and systematic sources of variation. MetaView software (version 5.3)[14] was used for all calculations. To investigate if the review was subject to publication bias, funnel plots were prepared by plotting the inverse of the standard error of the effect size estimate against the effect size.[15] These were examined for asymmetry. Where a study was found to report disparate results, sensitivity analysis was conducted to investigate the impact of that study on the findings.

Results

As shown in Figure 1, of the 803 papers located by the search strategy, 15 were included in the review.[5–9,16–25] These studies included 1340 participants ranging in age from 4 years 1 month to 18 years 11 months, and a relatively equal distribution of males and females (Table II). The majority of children with spina bifida had a lesion located in the lumbar or sacral region, used a device to assist with their mobility (e.g. a brace or wheelchair), and attended a mainstream school. However, children with spina bifida with a range of lesions and mobility levels were represented. Of the six studies[9,16,18,19,24,25] that reported the type of school attended by the participants with spina bifida, only four[9,18,19,24] stated the children were taught in a regular classroom; the other two studies[16,25] did not specify the type of classroom the participants were in. Seven studies[8,9,17,18,21,24,25] recorded the presence or absence of hydrocephalus or a shunt but none explored the effect of these variables on self-concept. The comparison group participants of typically developing peers were mostly matched for age and sex.

All studies examined self-concept as a multidimensional construct using a variety of self-concept measures (Table III). Six studies, however, only reported scores for the domain of global self-worth despite using a multidimensional scale.[5,18–20,22,25] Where contact details were provided, authors of these studies were contacted and supplementary information not included in their published manuscript was requested to assist with data analysis. Of the eight authors contacted, three responded, however, additional information was only available from one author (G Holmbeck).

The median quality assessment score was 6 out of 10 (range 1–10). Only four out of the 15 included studies specified their criteria for inclusion and exclusion for both children with spina bifida and typically developing peers (item 2). The two groups were comparable for all potential confounding factors in only five studies.

GLOBAL SELF-WORTH

Global self-worth is an overall judgement about how a child perceives their worth or value as a person (i.e. their self-esteem) including the extent to which they like themselves and are happy with the way they are leading their life.[25] Twelve studies compared the global self-worth of children with spina bifida with their typically developing peers. Ten studies concluded there was no difference between the groups for global self-worth (Fig. 2), while two studies found global self-worth was lower in children with spina bifida.[5,16] Meta-analysis revealed that global self-worth of children with spina bifida was significantly lower than children with typical

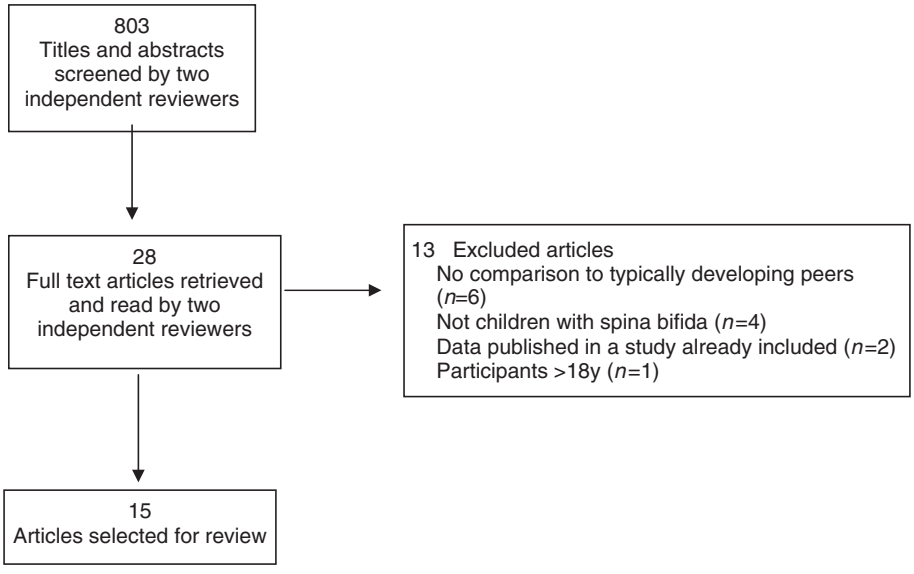

Figure 1: *Study selection process.*

Table II: Summary of included studies

Author	*QA /10*	*Sample size*	*Age*	*Sex*	*IQ*	*Schooling*	*Level of Lesion*	*Hydrocephalus/ shunt status*	*Mobility*	*Comparison group*	*Outcome measure*
Appleton et al.[9]	6	158 79 SB, 79 C	13y 7mo SB 13y 3mo C (7-18y 11mo)	76M 82 F 38M 41F in each group	78.9 (SD7.9) SB 100.8 (SD 14.9) C	49 SB mainstream, 17 special school, 13 other 63 C mainstream	Lumbar 26 Sacral 11 Thoracic 30 Cervical 3	70.9% had hydrocephalus 65.8% had surgery for shunt	No aids 16 Braces 31 Wheel chairs or carried 23	Matched for age (SD 6mo), sex, classroom & housing neighbourhood. No known chronic illness, disability or special educational needs	Self-perception Profile for Learning Disabled students (Renick & Harter[29])
Appleton et al.[18]	7	144 72 SB 72 C	14y SB (7-18y 11mo)	35M 37F	78.6 (SD 17.9) SB	45 mainstream, 14 special school, 13 other (SB)	Not reported	70.9% had hydrocephalus 65.8% had surgery for shunt	Not reported	Matched for age (SD 6mo), sex, classroom & housing neighbourhood. No known chronic illness, disability or special educational needs	Self-perception Profile for Learning Disabled students (Renick & Harter[29])
Campbell et al.[7]	5	40 20 SB 20 C	14y 4mo (SD 2y 5mo) SB 14y 3mo (SD 2y 5mo) C	8M and 12 F in each group	99.05 (SD 9.61) SB	Not reported	Lumbar 16 Sacral 4	Not reported	Not reported	Matched for age and sex	Tennessee Self-concept scale (Fitts[30])
Casari & Fantino[22]	6	248 40 SB 208 C	12y (SD 8–18y) SB Control group not stated	20M 20F SB 106M 102F C	70-120 (mean 90) for SB Controls not stated	Not reported	Not reported	Not reported	Not reported	Not reported	Perceived competence scale for children (Harter[26])
Edwards-Beckett[20]	6	60 30 SB 30 C	9y 6mo (SD 2y) SB C 9y 6mo (SD 1y 8mo)	12M 18F SB 15M 15F C	Children were in an appropriate school grade for their age	Not reported	Lumbo-sacral 52% Lumbar 19% Thoraco-lumbar 19% Sacral 10%	Not reported	Not reported	Not reported	Piers-Harris Self-Concept Scale for children (Piers[31])
Ellis[25]	10	62 31 SB 31 C	15.3 (13–17y 6mo) Ages of control group not stated	13M 18F in each group	>70	All attended mainstream schools (30 public, 1 private)	Lumbar 20 Sacral 10	67.7% had hydrocephalus No information on shunt status	No aids (14/31) Braces/ crutches (8/31) Wheelchair (9/31)	Matched for age, sex, ethnicity, socioeconomic status & type of school attended	Piers-Harris Self-Concept Scale for children (Piers[31]) Tennessee Self-concept Scale (Fitts[30])

Table II: (**Continued**)

Author	*QA /10*	*Sample size*	*Age*	*Sex*	*IQ*	*Schooling*	*Level of Lesion*	*Hydrocephalus/ shunt status*	*Mobility*	*Comparison group*	*Outcome measure*
Harvey & Greenway[16]	6	27 9 SB 18 C	Mean 10y 3mo for C group Not stated for SB group	8M 10F C Not stated for SB group	Intelligence within normal range	5 main-stream, 4 special school SB 18 main-stream C	6 myelomen-ingoceles 3 meningoceles	Not reported	Not reported	Recruitment through guiding and scouting movements. Children were selected by leaders of these organizations	Piers-Harris Self-Concept Scale for children (Piers[31])
Holmbeck et al.[8]	6	126 59 SB 67 C	8y 4mo (SD 6mo) SB 8y 6mo (SD 6mo) C (8–9y)	37M 31F in each group	PPVT-R scores 92.49 (SD 18.49) SB 108.97 (SD 15.06) C	Not reported	T/sp 13% L/sp 54% Sacral 32% 82% MMC 12% LMC 6% other	No information on hydrocephalus 71% had a shunt in situ	Mobile with no assistance 19% With braces 63% By wheelchair 18%	Recruited by contacting school where children with SB were enrolled	Self-perception Profile for Children (Harter[26])
Kazak & Clark[23]	6	62 30 SB 32 C	Not stated but < 18y	Not stated	Not reported	Not reported	Not reported	Not reported	Not reported	Recruited through local paediatricians, general paediatric clinics and a newspaper advertisement	Piers-Harris Self-Concept Scale for children (Piers[31])
Landry et al.[17]	7	30 15 SB 15 C	8y 2mo SB 8y 1mo C	9M 6F SB 6M 9F C	Intelligence within normal range	Not reported	Not reported	100% had hydrocephalus 100% had a shunt in situ	8 were ambulatory and 7 were non-ambulatory	Recruited from enrolment list of two community day care centres	Pictorial scale of perceived competence & acceptance for young children (Harter & Pike[32]) Self-perception Profile for Children (Harter[26])
MacBriar[24]	4	49 19 SB 30 C	12y (SD2.6) 5y SB 11.5 (SD 2). 13y C	9M 10F (SB) 11M 19F C	1 child diagnosed with intellectual disability Data not presented for 1 child	All children with two exceptions attended mainstream schools	Lumbar 3 Sacral 1 Don't know 15	57.9% had hydrocephalus 57.9% had a shunt in situ	Braces 12 (6 long leg, 6 short leg) Ambulatory 4 Non ambulatory 2	Siblings of the SB group were used as controls	Piers-Harris Self-Concept Scale for children (Piers[31])
Mobley et al.[6]	6	42 21 SB 21 C	64.7mo SB 64.3mo C	7M 14F in each group	Not reported	Not reported	All MMC	Not reported	All used mobility assistive devices with 67% using more than 1 device	Recruited through Spina Bifida Association (siblings of children with SB), day care centres and by personal contacts in the community	Pictorial Scale of Perceived Competence and Social Acceptance for Young Children (Harter & Pike[32])

Table II: (Continued)

Author	*QA /10*	*Sample size*	*Age*	*Sex*	*IQ*	*Schooling*	*Level of Lesion*	*Hydrocephalus/ shunt status*	*Mobility*	*Comparison group*	*Outcome measure*
Pearson et al.[5]	1	122 80SB 42C	'12 yr olds'	Not stated	Not stated	Not reported	Not reported	Not reported	Not reported	Not reported	Piers-Harris Self-Concept Scale for children (Piers[31])
Rodriguez Aponte[19]	9	40 20 SB 20 C	Mean age 10y 1mo (9–12y 5mo)	10M 10F in each group	Not stated but children matched for grade and grade point average	All attended mainstream schools	Moderate level of MMC	Not reported	All mobile with aid, 40% minimal usage, 60% use aid regularly	Three public schools in Puerto Rico Matched for age, sex, grade, socioeconomic status and grade point average	Piers-Harris Self-Concept Scale for children (Piers[31])
Thill et al.[21]	7	130 62 SB 68 C	8y 4mo (SD 6mo) SB 8y 6mo (SD 6mo)	37M 31F in each group	Not stated	Not reported	T/sp 13% L/sp 54% Sacral 32% 82% MMC 12% LMC 6% other	No information on hydrocephalus 71% had a shunt in situ	Mobile with no assistance 18% With braces 63% By wheelchair 19%	Recruited by contacting school where children with SB were enrolled	Self-perception Profile for Children (Harter[26])

QA; quality assessment; SB, spina bifida; C, comparisons; M, males; F. females; PPVT-R, Peabody Picture Vocabulary Test – Revised; T/sp, thoracic spine; L/sp, lumbar spine; MMC, myelomeningocele; LMC, lipomeningocele.

development with a medium-sized effect (d=−0.39, 95% CI −0.65 to −0.12; Fig. 2).

PHYSICAL APPEARANCE

The physical appearance domain explores how children feel about the way they look.[26] Data for the physical appearance domain of self-concept were reported in seven studies (Fig. 3). Five studies found no difference between children with spina bifida and their typically developing peers,[9,16,17,23,24] while two studies reported children with spina bifida scored lower on this domain than their typically developing peers. Meta-analysis revealed children with spina bifida scored significantly lower than children with typical development on the domain of physical appearance with a medium-sized effect (d=−0.26, 95% CI −0.46 to −0.06; Fig. 3).

ATHLETIC COMPETENCE

The athletic competence domain explores a child's perceived competence in their ability to participate in sports.[26] Four studies assessed the domain of athletic competence; all used a version of the Self-Perception Profile for Children.[6,9,17,21] Meta-analysis including three of these studies (Mobley et al.[6] did not report standard SDs for their cohort and so their data could not be included) found children with spina bifida scored lower than their peers with typical development on this domain with a medium-sized effect (d=−0.45, 95% CI −0.67 to −0.22; Fig. 3).

SOCIAL ACCEPTANCE

The social acceptance domain evaluates how a child feels about how they get along with their peers.[26] Four studies also assessed the domain of social acceptance using the Self-Perception Profile for Children.[6,9,17,21] Three studies concluded there was no difference between the groups for social acceptance (Fig. 3), while one study found social acceptance was lower in children with spina bifida.[9] Data from three of these studies were included in a meta-analysis (as Mobley et al.[6] did not report SDs for their cohort, their data could not be included) and results found scores for children with spina bifida overall were significantly lower for this domain compared with their peers with typical development with a medium effect size (d=0.33, 095% CI −0.55 to −0.11; Fig. 3).

Three studies[16,23,24] used the Piers-Harris questionnaire[31] which includes a domain related to social acceptance (popularity), however, overall there was no difference between the groups when a meta-analysis was conducted (d=−1.10, 95% CI −2.62 to 0.41; Fig. 3).

SCHOLASTIC COMPETENCE

The scholastic competence domain evaluates how children feel about how they perform at school.[26] Four studies examined the domain of scholastic competence using a version of the Self-Perception Profile for Children.[26] Three of these studies were included in a meta-analysis which found children with spina bifida had a lower self-concept than their typically developing peers with a medium-sized effect (d=−0.43, 95% CI −0.66 to −0.21; Fig. 3). The fourth study[6] also reported a significant difference between the groups (t=−2.8, p=0.008). Additionally, three studies assessed the related domain of intellectual competence and school status using the Piers-Harris Scale, which evaluates a child's

competence in schoolwork but also includes items relating to behaviour, anxiety, physical attributes, and popularity. These studies found no difference between children with spina bifida and children with typical development (Fig. 3).

BEHAVIOURAL CONDUCT

The behavioural conduct domain explores a child's perceived competence in how they behave.[26] There was no difference between the scores of children with spina bifida and their peers with typical development for any of the behavioural conduct domains (Fig. 4).

EFFECT OF AGE, SEX, AND SOCIOECONOMIC STATUS ON SELF-CONCEPT

There were insufficient data to perform meta-analysis on subgroups of participants according to age, sex, and socioeconomic status (as rated by Hollingshead Four Factor Index; Hollingshead AA, 1975, unpublished material). Two studies[9,18] found a main effect for sex on self-concept but another study[24] did not. Three studies[9,19,24] found no effect of age on self-concept, although another study[22] reported adolescents with spina bifida (aged 12–17y) scored lower on global self-worth than pre-adolescents with spina bifida (aged 8–11y; $p<0.01$). No difference was found between children with spina bifida and their typically developing peers from similar socioeconomic backgrounds for global self-worth, behavioural conduct, and social acceptance.[5,8] Children with spina bifida, however, scored lower on athletic competence and scholastic competence irrespective of socioeconomic background.[8]

SENSITIVITY ANALYSIS

Examination of the funnel plots found one study[16] appeared to have disparate results from the other included studies (Fig. 5). Sensitivity analyses were conducted to explore if removing the data from this study from the meta-analysis would affect the findings of the review. Excluding this trial did not change the results in the domains of global self-worth ($d=-0.31$, 95% CI −0.45 to −0.19); physical appearance ($d=-0.24$, 95% CI −0.49 to −0.002); behavioural conduct ($d=-0.18$, 95% CI −0.4 to 0.05); intellectual ($d=-0.24$, 95% CI −1.03 to 0.54; and happiness ($d=-0.35$, 95% CI −0.73 to 0.03). However, an effect for the popularity ($d=-0.41$, 95% CI −0.8 to −0.03) and anxiety $d=-0.7$, 95% CI −1.09 to −0.31) domains of the Piers-Harris scale was found when this trial was removed from the analysis. It is possible that these differences reflect the small sample size ($n=9$) of children with spina bifida in this study. It is also probable that selection bias was an issue in this study as the children from the comparison group with typical development were recruited through the guiding and scouting movements and were selected by leaders of these organizations to participate in the study.

Table III: Outcomes measures used to assess self-concept

Scale	*No of items*	*Description*	*Respondent*	*Example*	*Studies*
Piers-Harris Self-concept Scale for children[31]	80	Statements to which the child answers yes or no	Child	'I am well behaved at school'	Edwards-Beckett[20]; Ellis[25]; Harvey & Greenway[16]; Kazak and Clark[23]; MacBriar[24]; Pearson et al.[5]; Rodriguez[19]
Self-perception Profile for Children[26]	36	Each question presents two conflicting statements. One that most reflects the child is chosen and subsequently the child decides if it is sort of true or really true for them	Child	Some kids find it hard to make friends BUT other kids find it's pretty easy to make friends	Casari and Fantino[22]; Landry et al.[17]; Thill et al.[21]; Holmbeck et al.[8]
Self-perception Profile for learning disabled students[29]	46	Each question presents two conflicting statements. One that most reflects the child is chosen and subsequently the child decides if it is sort of true or really true for them	Child	Some kids read pretty fast BUT other kids are pretty slow readers	Appleton et al.[9]; Appleton et al.[18]
Pictorial Scale of Perceived Competence and Acceptance for young children[32]	24	Child looks at two pictures and is asked which of the children in the two pictures is most like him or her	Child	Questions are in pictorial format	Landry et al.[17]; Mobley et al.[6]
Tennessee Self-concept Scale[30]	100	Statements with five possible responses: completely false, mostly false, partly false-partly true, mostly true, completely true	Child	'I am a friendly person'*	Campbell et al.[7]; Ellis[25]

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Discussion

Meta-analysis revealed medium-sized (d=−0.26 to d=−0.45) and significant standardized mean differences to the effect that children with spina bifida scored significantly lower than their peers with typical development in the domains of global self-worth, physical appearance, athletic competence, social acceptance, and scholastic competence. It might not be unexpected for children with spina bifida to have a lower self-concept given that many of the characteristic symptoms associated with spina bifida such as incontinence, muscle paralysis, and musculoskeletal deformities may impact on their ability to function in their environment and interact with others. Indeed, children with spina bifida have been shown to be more dependent on adults, less likely to make independent decisions, have fewer social contacts outside of school, have lower scholastic abilities, and be less active than their typically developing peers.[8] Children with spina bifida often demonstrate poorer motor skills which can affect their play[17] and their ability to complete their activities of personal care.[18] This regular reminder of their physical disability and limitations might cue more frequent negative perceptions of their selves, resulting in frustration at their poorer motor skills and in less personal satisfaction in carrying out and completing tasks. These findings also show that they are aware they are different from their peers with typical development[9] and know that it is harder for them to take part in games and sports (athletic competence). Their lower self-concept in the social acceptance domain is consistent with previous literature that suggests children with disabilities often feel isolated from their peers.[9,27]

Although 10 of the 12 included studies reported no difference in the global self-worth of children with spina bifida and their peers with typical development, meta-analysis found a small but significant effect (d=−0.39) for this domain. Based on this effect size, a study would need to include at least 103 participants in each group to detect any difference in global self-worth between the groups. As none of the studies had included this number of participants, it would appear their sample sizes were too small to detect any overall difference. These results demonstrate the value of meta-analysis; the evidence from individual studies when viewed in isolation gave the impression of no difference; however, when the trends observed in these studies were aggregated, a significant difference was found.

The majority of participants with spina bifida attended mainstream schools, and are likely to have compared themselves to their typically developing peers rather than other children with spina bifida, which may reinforce the concept that they were different.[16,18] Children with spina bifida who attend mainstream schools place more importance on physical appearance, use social comparison processes to evaluate their own physical appearance, and feel less attractive when comparing themselves with their peers with typical development than to other children with physical disabilities.[9] These children are in a difficult position; they identify with their peers with typical development but may feel less competent and less accepted than those to whom they compare themselves.[9]

These results have implications for health professionals working with children with spina bifida who assist in the

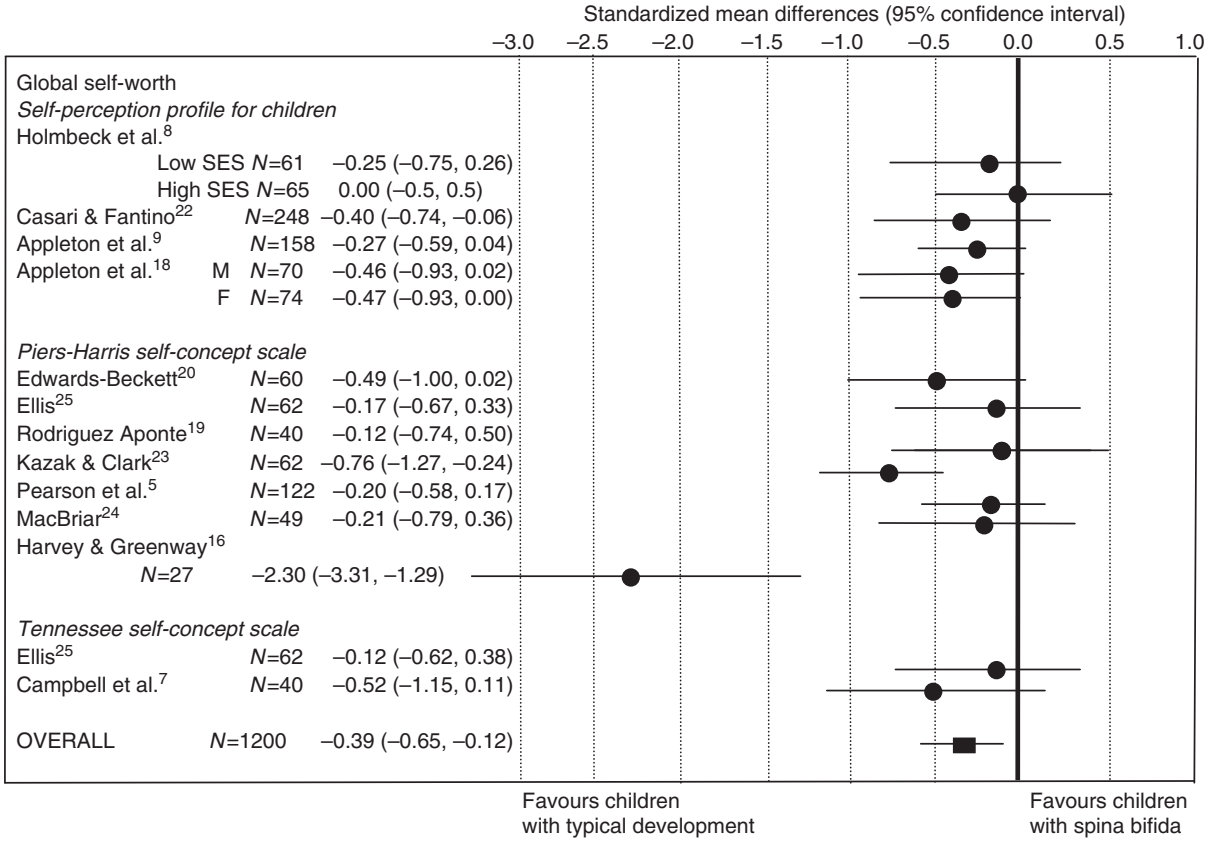

Figure 2: *Standardized mean differences and 95% confidence intervals for global self-worth.*

decision making processes about the management of a child's disabilities. Clinicians need to be aware that *on average*, children with spina bifida will have a lower self-concept; routine, and ongoing periodic measurement of self-concept in clinical practice, using an outcome measure with sound psychometric properties, such as the Self-Perception profile for children, would assist clinicians in being informed about how children with spina bifida feel about themselves. Such information would help clinicians identify children with spina bifida who have a lower self-concept, allowing them to work in partnership with the child to develop coping strategies.[9,24] Clinicians might help the child manage their negative body image through appropriate referral for cognitive and behavioural self-management interventions.[18] They might help negotiate child independence and autonomy,[18] e.g. helping them master those physical and cognitive skills they are capable of[6] and assist the child to discover development opportunities where they feel competent,[18] e.g. meeting other children with spina bifida or other physical disabilities perhaps through specialized camps or

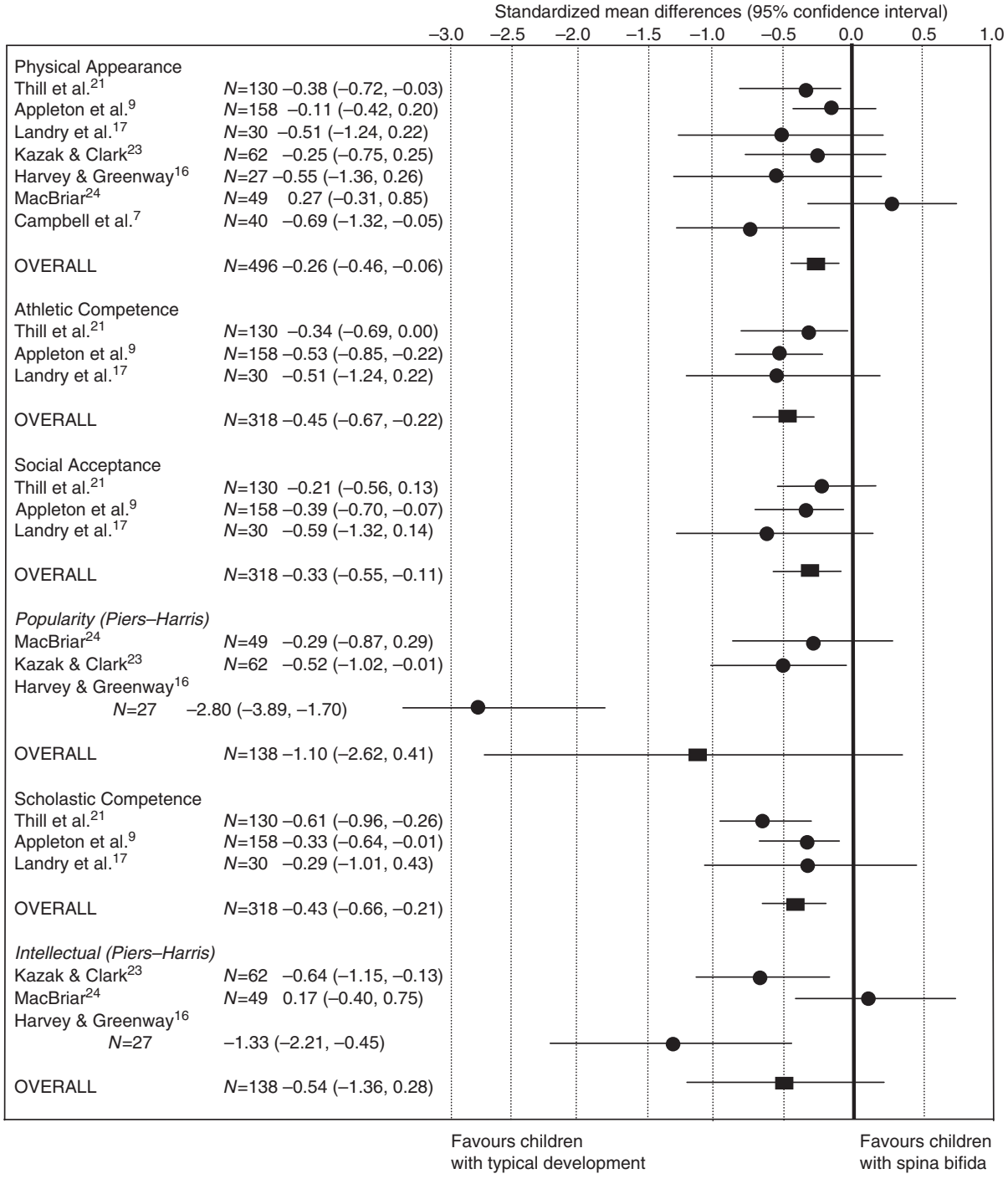

Figure 3: *Standardized mean differences and 95% confidence intervals for physical appearance, athletic competence, social acceptance and scholastic competence.*

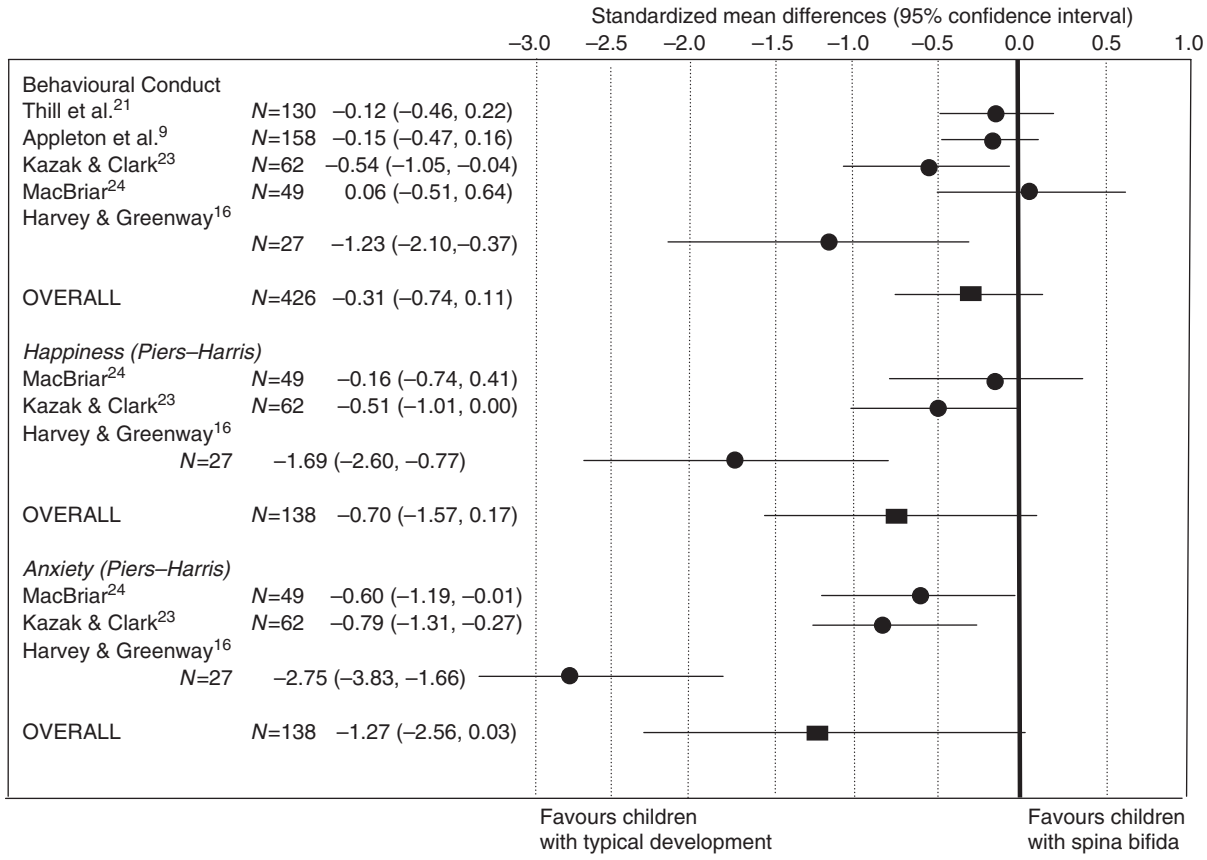

Figure 4: *Standardized mean differences and 95% confidence intervals for behavioural conduct.*

involvement in wheelchair sports. Such experiences may be valuable in helping children feel 'part of a peer group' or a larger community.[9]

It is important to remember, however, that the findings do not indicate that *all* children with spina bifida will have a low self-concept, only that this is the case on average. While it is useful for health professionals to know how an average group of children with spina bifida feel about themselves, in general they work with individual children and so cannot assume every child will have a low self-concept. Individual assessment, therefore, is very important when dealing with children who are unhappy with themselves, as this may not necessarily relate to their impairment, and so clinicians must be open to looking beyond the disability itself.

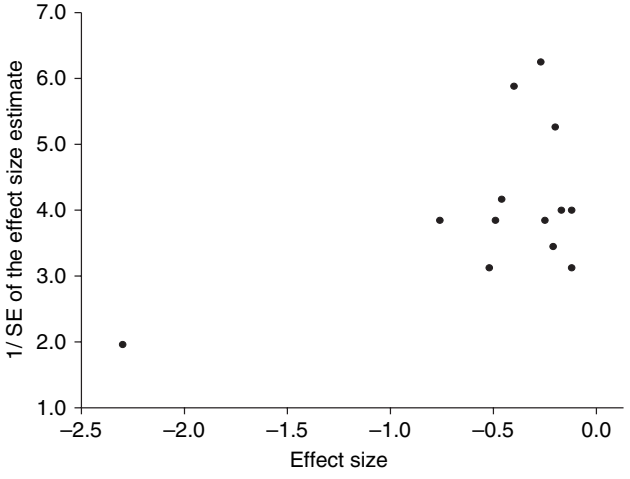

Figure 5: *Funnel plot to investigate for publication bias.*

The strengths of the review are that it used an extensive search strategy to locate 15 studies including 1340 participants and advanced the literature in this area by conducting a robust meta-analysis. Many of the included studies employed the same or similar outcome measures (e.g. Self-perception Profile for children and the Piers–Harris Self-concept Scale for children), making the results from the meta-analysis more robust. In a majority of cases the findings of these studies were consistent with each other. This review was limited in determining the effect of demographic characteristics such as age, sex, and socioeconomic status on self-concept in children with spina bifida because with only one exception, all the included trials did not report data for individual subgroups and so meta-analysis was not performed due to a lack of data. A further limitation is that although the random effects model assumed variability was due to a combination of random sampling error and systematic sources of variation, heterogeneity may still be an issue. Meta-analysis also cannot compensate for limitations in the original data such as the presence of uncontrolled confounding variables, measurement error, and selection bias.[28]

Conclusion

Children with spina bifida on average have a lower self-concept than their peers with typical development. Clinicians need to take account of this information in planning the assessment and treatment of this group. Finally, this review demonstrated the benefit of using meta-analysis to detect a moderate effect by improving the power of a number of small studies.

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