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PHYSIOLOGY & REHABILITATION | RESEARCH ARTICLE

Intellectual characteristics using WISC-IV in children with myelomeningocele

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Abstract: Purpose: Children with spina bifida are known to have various intellectual characteristics, including nonverbal learning disabilities due to hydrocephalus. While intelligence tests can ascertain the individual profiles of children with delays in intellectual development, there are few opportunities to clarify the intellectual characteristics of children who have never been identified as having delays in intellectual development. This study aims to clarify the intellectual characteristics as well as areas for intervention and improvement of children with myelomeningocele who show no delays in intellectual development. Methods: The Wechsler Intelligence Scale for Children—Fourth Edition was performed on six children (including four children with shunted hydrocephalus) aged 6–15 years. The Full-Scale Intelligence Quotient (FSIQ), index scores, and core subtest scores were analysed. Results: Processing Speed Index and Cognitive Proficiency Index were low. The General Ability Index was average, and there were no differences between Verbal Comprehension Index and Perceptual Reasoning Index. Conclusion: Children with myelomeningocele have low processing speeds. We consider the General Ability Index a better index of intrinsic intelligence than the FSIQ for such children. With appropriate support, children with spina bifida may be able to effectively demonstrate their skills and abilities at school and in society.

ABOUT THE AUTHORS

Hiroshi Mano and Nobuhiko Haga belong to the Department of Rehabilitation Medicine of the University of Tokyo Hospital. Their department focuses on clinical practice, research, and education of paediatric rehabilitation. The target diseases of research are rare and intractable diseases of children such as spina bifida, congenital limb deficiency, congenital insensitivity to pain with anhidrosis, fibrodysplasia ossificans progressiva, and skeletal dysplasias.

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PUBLIC INTEREST STATEMENT

Children with spina bifida are known to have various intellectual characteristics, including nonverbal learning disabilities, due to hydrocephalus. This study aims to clarify the intellectual characteristics as well as areas for intervention and improvement of children with myelomeningocele using Wechsler Intelligence Scale for Children—Fourth Edition. Children with myelomeningocele have low processing speeds and are limited in their abilities to effectively exhibit their core portion of intelligence. For children with spina bifida, the General Ability Index is a better index of intelligence than the Full-Scale Intelligence Quotient. We concluded that with appropriate support, children with spina bifida may be able to effectively demonstrate their skills and abilities at school and in society. Occupational therapy and neuropsychological rehabilitation may improve their abilities. When such children are performing tasks, it might be effective to extend deadlines and make the relevant considerations.

Subjects: Disability; Pediatrics & Child Health; Physiology

Keywords: spina bifida; myelomeningocele; intelligence; Wechsler scales; processing speed

1. Introduction

Spina bifida is defined as a congenital condition in which posterior elements of the spine (including spinous processes and vertebral arches) are defective. Spina bifida and anencephaly comprise neural tube defects. In spina bifida cystica, a protuberance is formed due to dorsal extrusion of the spinal or cauda equina nerve. Spina bifida occulta involves only dysraphism (incomplete or abnormal fusion) of spinal posterior elements with no protrusion of meninges or neural tissue. Spina bifida cystica is especially accompanied by abnormalities, including hydrocephalus, Chiari malformations, and syringomyelia. Frequently used classifications (Bartonek, Saraste, & Knutson, 1999) of neurological-level paralysis are (Sharrard, 1964) and (Hoffer, Feiwell, & Perry et al., 1973) (the latter differs from the Hoffer classification used for ambulation abilities; see hereafter). Meanwhile, Hoffer classification (Hoffer et al., 1973) is used to evaluate ambulation abilities.

The prevalence of spina bifida ranges from 3 to 8 per 10,000 births, showing differences by country and region. In Japan, the prevalence is 3.44 (Rosano et al., 1999). Although spina bifida can largely be prevented by folic acid intake (Viswanathan et al., 2017; MRC Vitamin Study research group, 1991), only 13.2% of preventable cases worldwide are actually prevented via folic acid intake (Arth, Kancherla, & Pachon et al., 2015).

Primary symptoms are motor and sensation disorders due to paralysis at the spinal and cauda equina nerve levels and excretory (urination and defecation) disorders. Secondary disorders can occur if temporary disorders are not properly managed. Central nervous system (CNS) disorders such as convulsions and intellectual disorders are preventable or controllable to a certain extent with appropriate management of hydrocephalus and Chiari malformations. Lower-limb deformations, mainly in the foot region, cause impairment of ambulation; treatments for these conditions include orthosis, surgery, and rehabilitation.

The characteristics of intellectual ability and cognitive functioning of children with spina bifida are variously known (Dennis, Landry, Barnes, & Fletcher, 2006). Such children generally have poor perception and mathematical abilities while linguistic abilities are preserved. However, there are deficits and strengths in specific abilities. For example, traditionally, these children are thought to be poor at perception tasks. Perception tasks in which children with spina bifida show relative weaknesses are figure-ground, drawings of geometric forms, and mazes; however, they have relative strengths in face perception and object perception (Dennis, Fletcher, Rogers, Hetherington, & Francis, 2002). As for language, while word decoding and word identification skills are preserved, reading comprehension, writing fluency, and discourse are considered as weak (Barnes & Dennis, 1992; Barnes, Dennis, & Hetherington, 2004; Barnes, Faulkner, & Dennis, 2001; Dennis & Barnes, 1993; Dennis, Jacennik, & Barnes, 1994). Other areas in which specific characteristics have been reported include mathematics (Barnes, Pengelly, & Dennis et al., 2002; Barnes, Stubbs, & Raghobar, 2011; Dennis & Barnes, 1993, 2002; English, Barnes, & Taylor et al., 2009; Raghobar, Barnes, & Dennis et al., 2015), attention (Brewer et al., 2001), and memory (Cull & Wyke, 1984; Yeates & Enrile, 2005; Yeates, Enrile, & Loss et al., 1995).

As for general intelligence, children with hydrocephaly (including spina bifida, cerebral aqueductal stenosis, and other conditions) are considered to have lower mathematical skills compared with reading (Barnes et al., 2002) and lower nonverbal intelligence compared with verbal intelligence (Brookshire, Fletcher, & Bohan et al., 1995; Dennis, Fitz, & Netley et al., 1981; Fletcher, Francis, & Thompson et al., 1992; Hetherington & Dennis, 1999). As for the scores on the Wechsler Intelligence Scale for Children (WISC) among children with spina bifida, Vinck et al. reported low

Performance Intelligence Quotient (PIQ) and Full-Scale Intelligence Quotient (FSIQ) on the WISC—Third Edition (WISC-III) among children with myelomeningocele (Vinck, Nijhuis-van der Sanden, & Roeleveld et al., 2010). Ito et al. reported that PIQ was significantly lower than Verbal Intelligence Quotient (VIQ) on the WISC—Revised (WISC-R) in children with spina bifida with hydrocephaly; they found that the extent of difference between these two intelligence quotients was correlated with enlargement of the posterior horn of the lateral ventricle and that lesion sites are in the visual pathway and visual cortex (Ito, Saijo, & Araki et al., 1997).

Wechsler scales are intelligence tests commonly used worldwide, and standardised Japanese versions are often used in Japan. For children, the newest version used in Japan is the WISC—Fourth Edition (WISC-IV) (Wechsler, 2010a, 2010b, 2014). The WISC-III newly introduced a General Ability Index (GAI) (Raiford, Weiss, & Rolfhus et al., 2005) and the WISC-IV a Cognitive Proficiency Index (CPI) (Weiss & Gabel, 2008). WISC-IV provides four index scores: Verbal Comprehension Index (VCI), Perceptual Reasoning Index (PRI), Working Memory Index (WMI), and Processing Speed Index (PSI). The GAI, which is calculated from the scores of VCI and PRI, provides the practitioner with a summary score that is less sensitive to the influence of working memory and processing speed than FSIQ. The CPI, which is calculated from the scores of WMI and PSI, represents a set of functions whose common element is the proficiency with which a person processes certain types of cognitive information. The FSIQ is calculated from all four index scores. Children with learning disabilities or attention deficit hyperactivity disorder (ADHD) have higher GAI than FSIQ in more than 70% of cases (Raiford et al., 2005). The GAI is reported to be higher than the CPI in children with brain tumours (Kahalley, Winter-Greenberg, & Stancel et al., 2016), learning disabilities (Cornoldi, Giofre, Orsini, & Pezzuti, 2014; Poletti, 2016), and ADHD (Devena & Watkins, 2012). To the best of our knowledge, WISC profile characteristics including the GAI and CPI in children with spina bifida are unknown.

Children with delays in intellectual development have opportunities to undergo intelligence tests to clarify their individual intellectual profiles. They may also engage in neuropsychological rehabilitation programmes in hospitals and educational support in their schools when necessary. However, children who seem to have no delays have no such opportunities. Considering the preceding study, we hypothesised that: (1) the PRI score of children with myelomeningocele is lower than the general population average; and (2) their PRI score is lower than their VCI scores. The purpose of this study is to clarify, using the WISC-IV, the intellectual characteristics of children as well as areas for intervention and improvement of children with myelomeningocele who have not displayed delays in intellectual development.

2. Materials and methods

2.1. Participants

The participants were recruited from outpatients of the Department of Rehabilitation Medicine of our university hospital or members of Shizuoka affiliate of Spina Bifida Association of Japan between September 2015 and February 2017. A total of six children with myelomeningocele ranging from 6 to 15 years of age who had never been identified as having delays in intellectual development were included. They were followed at the hospital specialised for children or the university hospital, and had never been examined using intelligence tests. The mean age was 9.8 years with a standard deviation of 3.8 years; there were four female and two male children. Four children had shunted hydrocephalus. Table 1 shows ambulation abilities and paralysis levels of each participant.

2.2. Procedure and measures

This study was approved by the Ethics Committee of the Faculty of Medicine of the University of Tokyo (approval number 10706). The study was explained to the children and their guardians, and their written consent was obtained, after which the WISC-IV was performed. The subtest scaled scores of 10 core subtests (standard scores of mean 10, standard deviation 3), FSIQ (standard scores of mean 100, standard deviation 15), and Index scores (standard scores of mean 100,

Table 1. Neurological level, ambulation and central nervous system complication of the participants

Child	Age (years)	Sex	Neurological level	Ambulation	Central nervous system complication
1	6.3	Male	Lower lumbar	Community ambulator	Hydrocephalus (treated with a shunt)
2	6.9	Female	Thoracic	Non-functional ambulator	Hydrocephalus (treated with a shunt)
3	7.0	Female	Sarcal	Community ambulator	None
4	9.8	Female	Sarcal	Community ambulator	None
5	13.2	Female	Lower lumbar	Community ambulator	Hydrocephalus (treated with a shunt)
6	15.6	Male	Lower lumbar	Household ambulator	Hydrocephalus (treated with a shunt)

standard deviation 15) were computed for each of the GAI, CPI, VCI, PRI, WMI, and PSI. From these scores, the intellectual characteristics of children with spina bifida were examined.

2.3. Statistical analysis

Wilcoxon signed-rank test was used for analysis under the hypothesis of mean 10 for subtest scaled scores and mean 100 for FSIQ and Index scores. To compare FSIQ and Index scores, Wilcoxon signed-rank test was used to determine differences in corresponding mean values. For statistical analysis, JMP® Pro 11.0.0 (SAS Institute, Tokyo, Japan) was used with statistical significance at $p < 0.05$.

3. Results

Table 2 shows each of the standard scores. Mean values were as follows: FSIQ 91.0, GAI 99.7, CPI 81.8, VCI 98.5, PRI 100.2, WMI 89.5, and PSI 79.3. The mean values of FSIQ, GAI, VCI, PRI and WMI were within one standard deviation (85–115), or average range. The mean values of CPI and PSI were within two standard deviations. Table 3 shows comparisons of standard scores. The CPI and PSI were significantly lower (together, $p = 0.03$) than 100, which was the population mean. The CPI was significantly lower than the GAI, and the GAI was significantly higher than the FSIQ (together, $p = 0.03$). Compared with the VCI, PRI, and WMI, the PSI was significantly lower (for each, $p = 0.03$).

4. Discussion

To the best of our knowledge, no WISC-IV profile characteristics, including the GAI and CPI, have been reported for children with spina bifida. In this study, we clarified these characteristics with the following points being salient:

- Processing Speed Index < Verbal Comprehension Index, Perceptual Reasoning Index, Working Memory Index;
- No significant differences exist between the Verbal Comprehension Index and Processing Speed Index;
- General Ability Index > Full-Scale Intelligence Quotient > Cognitive Proficiency Index.

(1) Processing Speed Index < Verbal Comprehension Index, Perceptual Reasoning Index, Working Memory Index:

Boyer et al. reported that children with myelomeningocele and shunted hydrocephalus have low working memories and processing speeds (Boyer, Yeates, & Enrile, 2006). In our study, while the

Table 2. Full-Scale Intelligence Quotient, index scores and subtest scale scores of WISC-IV

	Child 1	Child 2	Child 3	Child 4	Child 5	Child 6	Mean	Standard deviation	p-Value [§]
Full-Scale Intelligence Quotient (FSIQ) [†]	80	93	100	92	94	87	91.0	6.8	0.06
General Ability Index (GAI) [†]	94	105	110	96	99	94	99.7	6.5	0.81
Cognitive Proficiency Index (CPI) [†]	70	79	87	88	88	79	81.8	7.2	0.03
Verbal Comprehension Index (VCI) [†]	88	101	109	103	95	95	98.5	7.4	0.66
Similarities [‡]	5	12	11	10	10	10	9.7	2.4	1.00
Vocabulary [‡]	10	12	11	10	11	10	10.7	0.8	0.25
Comprehension [‡]	8	7	13	12	7	8	9.2	2.6	0.63
Perceptual Reasoning Index (PRI) [†]	95	109	109	89	104	95	100.2	8.4	0.94
Block design [‡]	9	14	9	10	9	9	10.0	2.0	0.75
Picture concepts [‡]	9	11	12	9	10	10	10.2	1.2	1.00
Matrix reasoning [‡]	10	9	13	6	13	9	10.0	2.7	1.00
Working Memory Index (WMI) [†]	79	85	91	91	100	91	89.5	7.0	0.06
Digit span [‡]	2	7	9	8	8	9	7.2	2.6	0.03
Letter-number sequencing [‡]	11	8	8	9	12	8	9.3	1.8	0.50
Processing Speed Index (PSI) [†]	70	78	86	88	81	73	79.3	7.1	0.03
Coding [‡]	2	4	7	8	5	4	5.0	2.2	0.03
Symbol search [‡]	7	8	8	8	8	6	7.5	0.8	0.03

Notes: WISC-IV: Wechsler Intelligence Scale for Children—Forth Edition.

[†] The FSIQ and the index scores have a mean of 100 and a standard deviation of 15.

[‡] The subtest scores have a mean of 10 and a standard deviation of 3.

[§] Wilcoxon signed-rank test.

Table 3. Pairwise comparisons of index scores of WISC-IV

	Mean	Standard error of the mean	p-Value [†]
GAI-FSIQ	+8.7	1.6	0.03
CPI-FSIQ	-9.2	1.6	0.03
GAI-CPI	+17.8	3.1	0.03
VCI-PRI	-1.7	3.5	0.88
VCI-WMI	+9.0	3.5	0.09
VCI-PSI	+19.2	1.7	0.03
PRI-WMI	+10.7	4.1	0.06
PRI-PSI	+20.8	4.2	0.03
WMI-PSI	+10.2	2.8	0.03

Notes: WISC-IV: Wechsler Intelligence Scale for Children—Forth Edition; FSIQ: Full-Scale Intelligence Quotient; GAI: General Ability Index; CPI: Cognitive Proficiency Index; VCI: Verbal Comprehension Index; PRI: Perceptual Reasoning Index; WMI: Working Memory Index; PSI: Processing Speed Index.

[†] Wilcoxon signed-rank test

WMI was not significantly low, we did confirm a significantly low PSI. It is necessary to consider the two possibilities with regard to intelligence: that processing speed itself is low and that, although processing speed itself is not low, it is decreased for some reason. One possible reason is upper-limb dysfunction. Patients with myelomeningocele display upper-limb dysfunction, and the dysfunction is related to a variety of primary or secondary conditions of spina bifida, including brain dysmorphologies, spinal malformations, and hydrocephalus (Dennis et al., 2009). Although no subject in this study identified upper limb dysfunction nor showed apparent upper-limb dysfunction at the time of the tests, subjects might have had slight upper-limb dysfunctions. This became obvious in the processing speed task performed on a desk. As an example of a relationship between upper-limb function and intelligence, fine motor skills are related to math fluency (Raghubar et al., 2015). If reliable standardised examinations for upper extremity function of children were available, we could have assessed them; however, those are not available, at least not in Japan. This is a future assessment question.

(2) No significant differences exist between the Verbal Comprehension Index and Processing Speed Index:

With editions of the WISC before the fourth, the PIQ and FSIQ were reported to be low for children with myelomeningocele (Vinck et al., 2010), and, for children with spina bifida with hydrocephalus, the PIQ was lower than the VIQ (Ito et al., 1997). Contrary to our expectation, our study results did not show low PRI or that the PRI was lower than the VCI. It is possible that this was due to revision of the WISC items and changes in its factor structure. In the WISC-III, with the two-factor structure of the VIQ and PIQ, the VIQ was comprised of the VCI and the Freedom from Distractibility Index (FDI, revised to WMI in the WISC-IV) while the PIQ was comprised of the Perceptual Organisation Index (POI, revised to PRI in the WISC-IV) and PSI. If low processing speed is a characteristic of children with spina bifida, then it is reasonable that the PIQ would be found low on the WISC-III and the PRI would not be low using the WISC-IV.

(3) General Ability Index > Full-Scale Intelligence Quotient > Cognitive Proficiency Index:

In this study, children with myelomeningocele had significantly low scores in both of the PSI core subtests and in one of the two WMI core subtests, resulting in a significantly low CPI. On the other hand, each score on the VCI and PRI core subtests was average, and, thus, the GAI was average. The GAI is an index related to the core portion of intelligence, and the CPI is an index of the fundamental skills indispensable for effectively exhibiting crystallised intelligence and fluid intelligence abilities. We

believe that, for children with spina bifida, the GAI is a better index of intelligence than the FSIQ. Children with spina bifida are limited in their abilities to effectively exhibit their core portion of intelligence; they require assistance in working memory and processing speed aspects to effectively demonstrate their abilities at school and in society. Special needs education, looking ahead to future social participation and employment, fulfils an important role. Occupational therapy and neuropsychological rehabilitation may also be effective in improving their abilities. Repeated exercise of tasks which require processing speed ability may strengthen their abilities. Preparation for such tasks in advance may be effective in demonstrating their abilities satisfactorily. On the other hand, a compensatory approach is also important. When such children are performing tasks, it might be effective to extend deadlines and to make the relevant considerations.

4.1. Limitations

There were only six participants in this study, which may have had an impact on the results. In the present study, we limited the participants to the patients without delays in intellectual development. Additionally, it was difficult to obtain cooperating participants for this study: as participants showed no apparent delays in intellectual development, there was no particular need for intelligence testing, and, at approximately 90 min, the duration of the WISC test is considered long. Although the sample size in this study was small, the participants were genuine targets as intended. Ambulation abilities, paralysis levels, and existence or non-existence of hydrocephalus varied among the children in this study. Whether or not the motor function, activities of daily living, and CNS complications influenced the characteristics of intelligence, is a future question.

With few participants, we could not analyse the effect of paralysis level or presence of hydrocephalus on WISC-IV scores. The severity of spina bifida symptoms is related to two aspects: paralysis level and the level of CNS complications. Future studies are needed to clarify the relationships between intelligence and paralysis level and the presence/absence of hydrocephalus.

In this study we only used the WISC as an intelligence test. Further investigations using other intellectual tests such as Kaufman Assessment Battery for Children and Das-Naglieri Cognitive Assessment System have possibilities of providing better understanding about the characteristics of intelligence. This study used the WISC for children and, thus, did not elucidate the intellectual profiles of infants or adults. Further investigations are needed on infants and adults.

5. Conclusions

Children with myelomeningocele have low processing speeds. We presume that, with the WISC, the GAI is a better index of intrinsic intelligence than the FSIQ for such children. With appropriate support, children with spina bifida may be able to demonstrate increased skills and abilities at school and in society.

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Competing interests

The authors declare no competing interests.

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