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Implications of family environment and language development: comparing typically developing children to those with spina bifida

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Abstract

Introduction This study examines the effect of family environment on language performance in children with myelomeningocele compared with age- and education-matched controls selected from the same geographic region.

Methods Seventy-five monolingual (English) speaking children with myelomeningocele [males: 30; ages: 7–16 years; mean age: 10 years 1 month, standard deviation (SD) 2 years 7 months] and 35 typically developing children (males: 16; ages 7–16 years; mean age: 10 years 9 months, SD 2 years 6 months) participated in the study. The Comprehensive Assessment of Spoken Language (CASL) and the Wechsler tests of intelligence were administered individually to all participants. The CASL measures four subsystems: lexical, syntactic, supralinguistic and pragmatic. Parents completed the Family Environment Scale (FES) questionnaire and provided background demographic information. Standard independent sample *t*-tests, chi-squared and Fisher's exact tests were used to make simple comparisons between groups for age, socio-economic status, gender and ethnicity. Spearman correlation coefficients were used to detect associations between language and FES data. Group differences for the language and FES scores were analysed with a multivariate analysis of variance at a *P*-value of 0.05.

Results For the myelomeningocele group, both Spearman correlation and partial correlation analyses revealed statistically significant positive relationships for the FES 'intellectual–cultural orientation' (ICO) variable and language performance in all subsystems ($P < 0.01$). For controls, positive associations were seen between: (1) ICO and lexical/semantic and syntactic subsystems; and (2) FES 'independence' and lexical/semantic and supralinguistic tasks.

Conclusions The relationship between language performance and family environment appears statistically and intuitively sound. As in our previous study, the positive link between family focus on intellectually and culturally enhancing activities and language performance among children with myelomeningocele and shunted hydrocephalus remains robust. Knowledge of this relationship should assist parents and professionals in supporting language development through activities within the natural learning environment.

Keywords
family environment
language, hydrocephalus, myelomeningocele, spina bifida

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Introduction

Children with myelomeningocele and shunted hydrocephalus (MMSH) commonly manifest language deficits that too often go unrecognized. These language differences are noted as early as pre-school years, when foundations are being formed, through adolescence, when pragmatic and higher-order language skills are applied daily in 'natural' environments such as home, school, medical clinics and community (Vachha & Adams 2002, 2003). Increasingly, early intervention models focus on provision of family-centred services in such natural learning environments (Majnemer 1998; Campbell *et al.* 2002; La Paro *et al.* 2004; Barnett & Hustedt 2005). The family's critical role in this natural learning environment varies, based on the individual family's perceived values and expectations of their child. Optimally, the child–parent interaction should create a milieu which cultivates the necessity for, and the benefits of, language. Previously we have described challenges related to temperament, memory, academic and language differences unique to children with MMSH. These differences distinguish these children from a broader category of children with chronic physical disabilities. The purpose of this report is to explore means by which families might be most supportive of natural learning environments to circumvent individual or collective challenges specific to children with MMSH.

Understanding the dynamic interactions between environmental contributors and the child's development related to functional skills (e.g. language) and participation in social roles continues to be a challenge. The Family Environment Scale (FES; Moos & Moos 1986) has been applied in studies describing family factors and their relationship to issues such as family stress or psychosocial adjustment in young adults with MMSH (Wallander *et al.* 1989; Holmbeck & Faier-Routman 1995; Loomis *et al.* 1997; Lemanek *et al.* 2000). In describing models for intervention, Dunst and colleagues (2005) has underscored the importance of the natural learning environment, incorporating concepts consistent with those captured in one of the FES personal growth dimensions – intellectual/cultural activities. In family settings that encourage learned helplessness, hypervigilant attention to achievement, and/or avoidance of leisure opportunities, the natural learning milieu for language development may suffer or disappear.

In an earlier study we provided the first description of the relationship between family environment and language performance in children with MMSH (Vachha & Adams 2005). In particular, we demonstrated that family involvement in intellectual and cultural activities was positively related to language performance in children with MMSH. This follow-up investigation examines the link between family environment and language performance, concentrating on families' relative priorities contributing to the child's 'personal growth': independence (IND), achievement orientation (AO), intellectual–cultural orientation (ICO) and recreation. Specifically, we sought to explore correlations between family emphasis on intellectual/cultural activities and language performance in a much larger cohort of children with MMSH matched to a group of geographically similar, non-affected children. Further, we examined differences in family environment priorities between families of the two participating groups (MMSH and controls).

Methods

Participants

Seventy-five children with MMSH [males: 30; ages: 7–16 years; mean age: 10 years 1 month, standard deviation (SD) 2 years 7 months] and their families were recruited consecutively from the tertiary level Spina Bifida Program at Texas Scottish Rite Hospital for Children (TSRHC), Dallas, Texas. Thirty-five typically developing children from the same geographical region (males: 16; ages 7–16 years; mean age: 10 years 9 months, SD 2 years 6 months) and their families served as controls. The Institutional Review Board of the University of Texas Southwestern Medical Center, Dallas, Texas and TSRHC approved research protocols and consent forms for this research effort.

To minimize confounding variables, strict criteria for inclusion and exclusion were utilized. Inclusion criteria – MMSH group: (1) diagnosis of myelomeningocele with Chiari II; (2) diagnosis of hydrocephalus <12 months of age; (3) placement of a ventriculo-peritoneal shunt <12 months of age; (4) seizures (if previously diagnosed) were well-controlled with no change in anticonvulsant medication within the prior 3 months. Inclusion criteria – MMSH and controls: (1) monolingual, English-speaking background; (2) full-scale intelligence scores (FSIQ) > 70 on the Wechsler scales of intelligence. Four children with MMSH initially referred for inclusion were omitted because of low FSIQ.

Exclusion criteria – MMSH: (1) *any* prior history of shunt infections; (2) history of shunt malfunction within prior 3 months. Exclusionary criteria – MMSH and controls: (1) uncorrected auditory or visual acuity deficits or motor disabilities precluding a pointing response; (2) prior diagnosis of mental retardation; attention deficit disorder; or recent history of clinical depression.

All 75 children with MMSH children attended mainstream schools at the time of study participation; 22 children were in regular classrooms with no special education services; 44 were in regular classrooms but received special education services (e.g. content mastery) as required, and nine children received most of their education through special education services. Six children had been referred for language therapy, but only four of the six received language services at the time of testing. All children in the control group were in mainstream classrooms and were reported by teachers as performing in the middle class quartiles in language arts/reading.

Among the MMSH group, 63 lived with both biological parents (in each case, the mother completed the questionnaire); two had grandfathers as primary caregivers; eight lived with a single parent (mother in seven cases); and two children were legally adopted (adoptive mothers completed the questionnaire). Twenty-nine children in the control group lived with both biological parents, one was legally adopted, and five lived with a single parent (mother in all cases). Questionnaires were completed by mothers for all 35 controls.

Measures

Primary caregivers in both MMSH and control groups completed the FES, a 90-item true/false self-report instrument designed to gather information of family functioning on subscales including cohesion, expressiveness, conflict, IND, AO, ICO, moral–religious emphasis, organization and control. The format of the FES allows the family to rate their perceived emphasis on each of the above areas as either 'true most of the time' or 'false most of the time'. For the purpose of this study, *T*-scores (mean 50, SD 10) were derived for four subscales: IND, AO, ICO and active recreational orientation (ARO). Table 1 provides descriptions of the FES scales used. Socio-economic status (SES) was measured by the Hollingshead's Four Factor Index (Hollingshead 1975) using four factors for computation: marital status, gender, occupation and education.

Subtests of the Comprehensive Assessment of Spoken Language (CASL; Carrow-Woolfolk 1999) were administered to all participants individually, and standard scores (mean 100, SD 15) were computed within lexical/semantic, syntactic, supralinguistic and pragmatic language subsystems. Table 1 provides definitions of language subsystems.

General cognitive ability (FSIQ) was determined by either the Wechsler Abbreviated Scale of Intelligence (WASI; Psychological Corporation 1999) or the Wechsler Intelligence Scales for Children – Fourth Edition (WISC-IV; Psychological Corporation 2003). The corrected correlation coefficient between the FSIQ composites measured by the two instruments is 0.86, suggesting that they measure highly similar constructs. Non-verbal intelligence was determined by the Performance IQ score on the WASI or the Perceptual Reasoning Index Score on the WISC-IV; corrected correlation coefficient between the non-verbal intelligence composites measured by the two instruments is 0.78. Verbal intelligence was also determined by either the WASI or WISC-IV with corrected correlation coefficient between the verbal intelligence composites measured by the two instruments reported as 0.84.

Statistical analyses

SAS (version 9.1) was used for data analyses. Standard independent sample *t*-tests, chi-square and Fisher's exact tests were used to make simple comparisons between groups for age, SES, gender and ethnicity. Spearman correlation coefficients were used to detect associations between language and FES data. Group differences for the language and FES scores were analysed with a multivariate analysis of variance (MANOVA) at a *P*-value of 0.05. *Post hoc* multiple comparisons (Bonferroni-corrected) were used to determine group differences within

Table 1. Descriptions of language and family environment categories

Category	Description
Language	
Lexical/semantic	Knowledge and use of words
Syntactic	Knowledge and use of grammar
Supralinguistic	Understanding non-literal language, ambiguities and inferences
Pragmatic	Functional use of language in social situations
Family environment	
Independence	The extent to which the family members are encouraged to be self-sufficient
Achievement orientation	The extent to which activities (school and work) are cast into an achievement-oriented or competitive framework within the family
Intellectual–cultural orientation	The level of interest in intellectual and cultural activities within the family
Active recreational orientation	Amount of participation in social and recreational activities

each of the language and FES categories such that *P*-values less than 0.013 and 0.01 were considered statistically significant for the FES and language scores, respectively.

Table 2. Descriptive statistics

	MMSH ($n = 75$)	Controls ($n = 35$)
Age, mean (SD)	10 years 1 month (2 years 7 months)	10 years 9 months (2 years 6 months)
Gender, *n* (%)		
Males	30 (40)	16 (46)
Females	45 (60)	19 (54)
Ethnicity *n* (%)		
Caucasian	57 (76)	25 (71.4)
Hispanic	9 (12)	6 (17.1)
African American	5 (6.7)	3 (8.6)
Other	4 (5.3)	1 (2.9)
Lesion level *n* (%)		
Thoracolumbar	7 (9.3)	
Mid-lumbar	53 (70.7)	
Lumbosacral	15 (20)	
Variable, mean (SD)		
Verbal intelligence	96.3 (13.8)	107.7 (13)
Non-verbal intelligence	89.3 (10.3)	109.1 (10.1)
Full-scale intelligence	91.3 (11.7)	104.8 (10.8)
CASL lexical/semantic	92.4 (13.4)	112.4 (16.5)
CASL syntactic	93 (10.6)	108.3 (12.5)
CASL supralinguistic	84.6 (10.5)	109.7 (8.1)
CASL pragmatic	82.7 (11.2)	104.8 (9.4)
FES independence	46.9 (9.4)	46.7 (8.8)
FES achievement orientation	47.6 (9.2)	48.1 (9.8)
FES intellectual–cultural orientation	48.9 (11.9)	56.7 (13.1)
FES active recreational orientation	48.8 (10.6)	54.3 (12.8)
SES	42.7 (11.5)	46.7 (11.6)

CASL, Comprehensive Assessment of Spoken Language; FES, Family Environment Scale; MMSH, myelomeningocele and shunted hydrocephalus; SD, standard deviation. Intelligence and Language scores reported as standard scores (mean 100, SD 15); FES scores reported as *T*-scores (mean 50, SD 10).

Results

Descriptive statistics

Descriptive statistics for the study samples are summarized in Table 2. On average, children with MMSH in this study showed verbal, non-verbal and full-scale intelligence within the normal range. Sample means for the FES scores were within one standard deviation of the normative mean of 50. SES of the caregivers in both groups ranged from professional to unskilled, representative of both high-income and low-income backgrounds, but the majority were in the middle-income group. Preliminary analyses showed no significant group differences in age ($t_{[108]} = -1.63$, $P = 0.11$), SES ($t_{[108]} = -1.7$, $P = 0.09$); gender ($\chi^2_{[1]} = 0.32$, $P = 0.57$); or ethnicity ($P = 0.81$, Fisher's exact test).

Correlations between family environment and language performance

For the MMSH group, Spearman correlations revealed statistically significant positive relationships for the ICO variable and performances within each of the four language subsystems (Table 3). The other family variables (IND, AO and ARO) showed no correlation with performance within any of the language subsystems ($P > 0.05$). No correlations were noted between age, lesion level, ethnicity and gender differences, and their relationship to family environment and language variables.

For the control group, Spearman correlations revealed statistically significant positive relationships between ICO and lexical/semantic and syntactic subsystems. Focus of the family on IND was related with increased performance in lexical/semantic and supralinguistic language subsystems (Table 3).

As effects of ICO on language outcomes may be confounded by the child's intelligence, we examined the correlations between ICO and each of the language subsystems after con-

Table 3. Spearman correlation analyses: FES, SES factors and language subsystems

	MMSH ($n = 75$)					Controls ($n = 35$)				
	IND	AO	ICO	ARO	SES	IND	AO	ICO	ARO	SES
LEX	0.03	0.14	0.38**	–0.04	0.34**	0.52**	0.08	0.34*	0.12	0.41**
SYN	–0.10	0.10	0.42**	–0.06	0.26*	0.26	0.17	0.41**	0.01	0.35*
SUPR	0.02	0.13	0.36**	0.01	0.28*	0.47**	0.11	0.29	0.04	0.32*
PRAG	0.11	0.10	0.37**	0.14	0.30**	0.20	0.08	0.06	0.27	0.29

FES, Family Environment Scale; SES, socio-economic status; MMSH, myelomeningocele and shunted hydrocephalus; IND, FES independence score; AO, FES achievement orientation score; ICO, FES intellectual–cultural orientation score; ARO, FES active recreation orientation score; LEX, lexical/semantic score; SYN, syntactic score; SUPR, supralinguistic score; PRAG, pragmatic score.
*$P < 0.05$, **$P < 0.01$.

Table 4. Partial correlations between language variables and ICO controlling for verbal and non-verbal intelligence

		LEX	SYN	SUPR	PRAG
MMSH	ICO (partial verbal intelligence)	0.12	0.21	0.13	0.18
	ICO (partial non-verbal intelligence)	0.32**	0.37**	0.31**	0.34**
Controls	ICO (partial verbal intelligence)	0.03	0.22	0.10	0.10
	ICO (partial non-verbal intelligence)	0.29	0.35*	0.26	0.07

ICO, intellectual–cultural orientation; MMSH, myelomeningocele and shunted hydrocephalus; LEX, lexical/semantic score; SYN, syntactic score; SUPR, supralinguistic score; PRAG, pragmatic score.
*$P < 0.05$, **$P < 0.01$.

Table 5. Partial correlations controlling for ICO and SES

		LEX	SYN	SUPR	PRAG
MMSH	SES (partial ICO)	0.17	0.04	0.12	0.14
	ICO (partial SES)	0.26*	0.35**	0.26*	0.26*
Controls	SES (partial ICO)	0.31	0.21	0.22	0.29
	ICO (partial SES)	0.20	0.31	0.19	–0.06

ICO, intellectual–cultural orientation; SES, socio-economic status; LEX, lexical/semantic score; SYN, syntactic score; SUPR, supralinguistic score; PRAG, pragmatic score; MMSH, myelomeningocele and shunted hydrocephalus.
*$P < 0.05$, **$P < 0.01$.

trolling for verbal and non-verbal intelligence for each of the two groups. For the MMSH group, significant correlations remained between each of the language subsystems and the ICO factor after controlling for non-verbal intelligence scores. Controlling for verbal intelligence scores, the estimated partial correlations between each of the language subsystems and the ICO factor no longer remained significant. Findings suggest that in the MMSH group the effect of ICO on language may be associated with the child's verbal (but not non-verbal) intelligence. Table 4 illustrates these findings.

For the control group, the estimated partial correlations between each of the language subsystems and the ICO factor revealed no significant correlations with all four language subsystems after controlling for verbal intelligence. Controlling for non-verbal intelligence revealed significant correlations only between ICO and syntax in controls (Table 4).

Correlations between SES and language performance

For both groups, there was an initial relationship that suggested SES correlated positively with language performance for each language subsystem (Table 3). However, as ICO and SES were themselves positively correlated ($rho_{MMSH} = 0.53$, $P < 0.01$; $rho_{controls} = 0.43$, $P < 0.01$), controlling for the ICO factor, the estimated partial correlation between each of the language subsystems and SES revealed no significant correlations for either the MMSH or control groups ($P > 0.05$). For completeness, we also controlled for SES and found significant correlations remained between ICO and each of the language domains for the MMSH group ($P < 0.05$); among the control group, no similar significant correlations were found ($P > 0.05$). This suggests that SES did not add anything unique to the language performance in children with MMSH. Table 5 illustrates these findings.

Group differences – language

Multivariate analysis of variance revealed significant group differences for the language variables (Wilks Lambda = 0.36, $P < 0.01$). Univariate analyses of variance (ANOVA) identified significantly poorer performance on average by the MMSH group in comparison with controls within each of the four language subsystems ($P < 0.01$).

Group differences – family environment

The 'family environment' MANOVA was also significant (Wilks Lambda = 0.9, $P = 0.03$). ANOVA revealed significant group differences for the ICO variable with the MMSH group scores falling below the controls ($F_{1,108} = 9.63$, $P = 0.0024$). Although significant group differences were noted for the ARO variable ($F_{1,108} = 5.53$, $P = 0.021$), using Bonferroni correction this was no longer significant. Univariate analyses for the remaining family environment parameters failed to reach statistical significance.

To test the possibility that cognition may mediate associations between group status and the ICO variable, the ANOVA was re-run with verbal and non-verbal intelligence as covariates. Group differences for the ICO variable became non-significant ($P > 0.05$), indicating that the *perception* of cognition and *responses to those perceptions* may account for group differences.

Discussion

This study explored the relationship between family environmental factors and language performance among children with MMSH in comparison with a non-affected matched cohort. The results in this extended study endorsed the findings in our original pilot report – family emphasis on activities identified in the FES category 'intellectual and cultural orientation' was positively related to language performance within all four language subsystems for children with MMSH. This was noted in each case, irrespective of ethnic background.

The difference in language performance scores comparing the MMSH group with controls was statistically significant and not unexpected; lower scores were noted in all four CASL language subsystems among those with MMSH. The striking difference came in the patterns of reported family priorities as described on the FES personal growth dimensions. The families of children with MMSH were significantly different from families of their matched controls in that, as a group, there was notably less emphasis reported on intellectual/cultural activities within the households.

Among the control group, different associations between language performance and family environment were noted. ICO was related to only two language subtests, lexical/semantics and syntax. Unlike the MMSH group, controls showed positive associations between FES-IND and lexical/semantic and supralinguistic performance. This seems reasonable given that typically developing children often experience fewer barriers to their natural exploration and independent actions. Further, with higher overall scores on language skills at baseline, different dimensions of family environment may play a role in projecting further language development.

The effect of cognition on language is important in interpreting these statistical data and in considering how parental perceptions of their child's intelligence might affect the dynamics of family environment processes. Controlling for the effect of verbal intelligence resulted in no significant correlations between ICO and any of the language subsystems in children with MMSH. On the other hand when controlling for non-verbal intelligence, significant correlations between ICO and language subsystems remained robust for the MMSH group, implying verbal (but not non-verbal) intelligence may have mediated associations between these variables. Language outcomes in children with MMSH may reflect an interactive cause rather than a primary consequence of the environment, mediated in part by families regulating intellectual cultural activities in accordance with what they perceived (correctly or incorrectly) as their child's intelligence/language abilities, and their child's ability to plan and follow through with ICO-related tasks.

When the developmental learning environment and the parental understanding of the child's cognitive abilities are in sync, this 'goodness-of-fit' in the parent–child dyad should serve to facilitate optimal language development. One would expect that for parents who accurately recognize the cognitive capabilities of their child and tailor the learning environment accordingly, the resultant scaffolding of supports might encourage language development. The strong association of ICO with language in MMSH seemingly supports the concept.

Alternatively, if strong emphasis is placed on 'pushing' the child to demonstrate tasks out of the contextual milieu (more reflected by AO on the FES), this might impede the natural learning process. Likewise for recreational orientation, the question arises: does the family perceive the role for the child as that of observer, or is it one of 'active participant' – thereby providing a natural learning environment in which to practice functional language skills?

Family influences on children's functions, activities and participation have become increasingly recognized as salient modifiers on developmental trajectories. In the literature on both myelomeningocele and the non-disabled populations, this mediating effect of family environment is dynamic, significant, and worthy of better understanding (Girouard *et al.* 1998; McKernon *et al.* 2001; Holmbeck *et al.* 2002; Friedman *et al.* 2004; McCarty *et al.* 2005).

Acquisition and application of language are central to all developmental strands. In considering the potential opportunities for family environment to enhance development, Johnson (2005) provides a critical distinction between two dimensions of family life: family process and family structure. In many studies published on effects of 'family environment' the concept is structural: marital status, number of household members, economic indicators and the like (Wallander *et al.* 1989). Clearly, these variables can impact developmental trajectories.

In the present study, aspects of family structure, including SES and ethnic heritage, did not make unique contributions to the measured language performance in the larger MMSH group. Nor did they reach significance when comparing the

MMSH to control participants. A wide range of factors including the qualitative nature of the time spent in daily interactions may well impact language more than those structural variables such as SES.

Accordingly, this study is more aligned with aspects of family process concerned with quality, direction and the valence of interactions, particularly between the family and child. The FES was selected in this study in an attempt to gather better descriptions of real-life exposures relative to intellectual pursuits and their relations to language. Wright and colleagues (2001) offer an intriguing example of the necessity to 'drill down' into such process questions when describing the mediating effects of a variable (television watching) to developmental outcomes. In a longitudinal, cohort comparison study of economically at-risk pre-school children, not only was the total time of television watching monitored, but the content and intellectual nature of the programming was also noted. Among those with greater viewing of exploratory content (compared with 'entertaining' content) a relationship was found related to greater language skills and increased exploratory play.

The value of developmental surveillance compared with cross-sectional screening underscores the importance of assessing children over time to gather better understanding of their developmental trajectories. Families clearly go through developmental changes as well. Unless given sequentially and longitudinally, the FES would not be expected to reflect those changes and relate them to the child's age/developmental progression. For both groups in this study, family processes different from those described herein may have played modifying roles in the development of language skill. As with many questionnaires, the FES provides a description of the represented family with the element of family projections to be considered. Although the FES does not provide real-time observations of the parent–child dyad, a limitation of this study, our results do offer important considerations for family intervention strategies.

Dunst and colleagues (2005) described a model for the natural learning environment distinguishing between those settings that are contextually (rather than non-contextually) based, and that are child directed (rather than adult directed) in nature. Family processes captured within the FES dimension of ICO are conceptually consistent with Dunst's optimal natural learning environments wherein 'learning opportunities [are] provided in everyday activity settings . . . when the learning itself is contextualized, functional, and socially adaptive' (Dunst *et al.* 2005). Emerging professional practices incorporating natural learning environments have evolved and show promise for enhanced parent–child interaction and child adaptation.

Conclusion

This study of language performance among children with MMSH offers insight into the mediator effect of the family environment process. The link between family focus on intellectually and culturally enhancing activities and language performance among children with MMSH remains robust. In offering suggestions to parents and programmes involved in early developmental intervention for children with MMSH, the words of Farver (1999) seem salient in light of this study: the optimal milieu for enhancing language skills is 'made up of everyday experiences rather than a deliberate curriculum, and they contain ordinary settings in which children's social interaction and behaviour occurs. They are the who, what, where, when and why of daily life.'

Key messages

- Children with myelomeningocele and shunted hydrocephalus (MMSH) demonstrate language difficulties despite average intelligence.
- Language deficits affect not only academic performance, but also the ability of the child to communicate effectively in community and medical settings.
- Family influences on children's functions, activities and participation have become increasingly recognized as salient modifiers of developmental trajectories.
- The link between family focus on intellectually and culturally enhancing activities and improved language performance among children with MMSH was clearly demonstrated in this study.
- In the literature on both myelomeningocele and the non-disabled populations, this mediating effect of family environment is dynamic, significant, and worthy of better understanding as we try to develop and implement early intervention programmes for children with developmental differences.

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