Condition-Related Knowledge Among Children with Spina Bifida: Longitudinal Changes and Predictors

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Objective To examine changes in three domains of condition-related knowledge among youth with spina bifida and to examine the utility of youth cognitive ability level and condition severity as predictors of knowledge change. **Methods** Seventy preadolescents with spina bifida completed a 12-item questionnaire assessing knowledge of spina bifida at three time points during middle childhood and early adolescence. Specific domains of knowledge assessed included (a) etiology of spina bifida, (b) functional status, and (c) shunt functioning (completed by participants with shunted hydrocephalus only). **Results** Findings revealed gains in accuracy of knowledge on 6 of 12 items; however, neither children's cognitive ability level nor condition severity predicted changes in knowledge over time. Most condition domains were characterized by low-to-moderate levels of knowledge across time. **Conclusions** Although significant gains were evident in children's condition-related knowledge, at Time 3, many participants still failed to understand basic information about the etiology of their condition or major functional issues associated with spina bifida. Additional education about catheterization and shunt malfunction are two domains that may be of particular clinical significance.

Key words adolescence; chronic illness; condition knowledge; spina bifida; understanding of condition.

Spina bifida (SB) is one of the most common birth defects, with an incidence rate of 1 in every 1000 live births (Charney, 1992). It is characterized by an incomplete development of the spinal column, resulting in neurological insult below the lesion site. Associated impairments in ambulation and bowel and bladder functioning are common, as are neuropsychological difficulties secondary to brain malformations and hydrocephalus. The severity of SB varies as a function of lesion level and neurological status; however, most children with SB experience significant functional impairment. As a result, children and their families must manage complex medical regimens including catheterization, bowel programs, use of orthopedic devices, skin checks to avoid pressure sores, and monitoring of shunt functioning, while navigating the normal developmental tasks of childhood and adolescence (Charney, 1992).

The examination of condition-related knowledge among youth with chronic medical and neurological conditions is important for several reasons. First, an understanding of children's condition-related knowledge has significant implications for the provision of clinical services. Children who fail to recognize warning signs of problematic functioning in one or more condition-related domains (e.g., signs of shunt malfunctioning and symptoms of urinary tract infections) may delay seeking treatment and may be at risk for greater medical morbidity. Assessing knowledge during the adolescent developmental period may be of particular importance as this is generally a period when increased regimen responsibilities

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are gradually transferred from parent to child (Drotar & Ievers, 1994). Thus, identifying areas in which preadolescents and adolescents lack sufficient condition-related knowledge can inform clinical education efforts. In the same vein, the evaluation of children's knowledge across multiple content areas can elucidate aspects of their condition for which knowledge is less well developed, thereby increasing the ability of parents and health professionals to address these issues. Second, prior research has indicated that children's knowledge of their own medical condition may affect not only how they conceptualize their condition, but also their ability to cope with it. Condition-related knowledge can decrease distress and anxiety and increase children's self-efficacy and adherence to medical regimen (Bartholomew et al., 1991; Edwinson, Arnbjornsson, & Ekman, 1988; Ievers et al., 1999; Lorenz, Christensen, & Pichert, 1985). Thus, knowledge about one's medical condition may have implications for psychosocial adjustment.

Although a lack of knowledge in one or more of the above domains could impair one's ability to manage his or her medical regimen, acquisition of condition-related knowledge is a topic that has been rarely studied in pediatric populations, including youth with SB (Wills, Burk, & Hayes, 1993). Among youth with other medical conditions, some past research has investigated the stages of development of condition-related knowledge, whereas other research has attempted to document the content of children's knowledge (Celando, Geller, Phillips, & Ziman, 1998; Cromer et al., 1990; Johnson, 1984). Research investigating the development of condition-related knowledge in both healthy children and children with chronic illnesses has established that one's understanding of medical illness and health progresses in accord with Piaget's theory of cognitive development (Berry, Hayford, Ross, Pachman, & Lavigne, 1993; Feldman & Varni, 1985; Perrin & Gerrity, 1981). In general, preadolescents tend to conceptualize illness in external, global, and nonspecific terms, have difficulty distinguishing symptoms from causes, and are less able to understand abstract illness concepts (e.g., prevention). In contrast, adolescents tend to conceptualize illness internally, according to specific symptoms, and can better understand concepts of causation (e.g., germs; Berry et al., 1993; Brewster, 1982; La Greca, Follansbee, & Skyler, 1990; Perrin & Gerrity, 1981).

In addition to focusing on the development of children's condition-related knowledge, past research has also investigated the content of children's knowledge at different ages or developmental stages. Collectively, these studies suggest that deficits in children's understanding of specific domains of their condition are common and that these deficits relate to condition management (Celando et al., 1998; Ievers et al., 1999). Interestingly, knowledge across different condition-specific content areas is often not related, emphasizing the need to assess multiple domains of knowledge (Johnson, 1984). Among youth with SB, findings suggest higher levels of knowledge for the functional aspects of the condition and deficiencies in knowledge related to diagnosis (type of SB and lesion level) and sexual functioning (Cromer et al., 1990; Erickson, 1992; Erickson & Erickson, 1992).

Although some prior research has investigated condition-related knowledge, past research is characterized by several deficits including: (a) a failure to consider the influence of potential confounding variables such as condition severity or cognitive ability level in predicting children's knowledge, (b) the use of cross-sectional research designs that include a large age range of participants, practices which do not allow for an examination of the developmental trajectory of condition-related knowledge, (c) a failure to include key demographic information about the sample under investigation, thus limiting the generalizability of findings, and (d) the use of mixed illness samples with small sample sizes of children within specific illness groups, thereby precluding analyses based on condition type or severity.

The current study builds on prior research by investigating the development of specific domains of condition-related knowledge among youth with SB. The current study utilized a longitudinal approach to investigate changes in three broad domains of children's knowledge across middle childhood and early adolescence: (a) knowledge of the etiology of SB, (b) knowledge of the functional status of individuals with SB, and (c) knowledge of shunt functioning among children with shunted hydrocephalus. We expected that significant increases in accuracy of children's knowledge in each of the above domains would occur during middle childhood and early adolescence such that by early adolescence, most youth would evidence an accurate understanding of the etiology of SB and the functional status of individuals with SB. Similarly, we expected that among those children with shunts, the majority would be able to provide accurate information about the purpose of a shunt, signs of shunt malfunction, and treatments for shunt-related problems by early adolescence.

Because children's knowledge of their medical condition may vary according to their cognitive ability level, as well as the severity of their condition and the complexity of the associated medical regimen, a secondary purpose of this study was to examine predictors of

changes in condition-related knowledge across the early adolescent developmental period. We predicted that cognitive ability level would be positively associated with increases in knowledge over time. With respect to condition severity, two competing hypotheses were proposed. First, given that greater condition severity may be associated with more neuropsychological impairments and that neuropsychological impairments may have a negative impact on the acquisition of condition-related knowledge, the first hypothesis proposed a negative relationship between condition severity and development of knowledge. The alternative hypothesis proposed that because children with more severe impairments have more experience in the daily management of condition-related issues, they may have higher levels of knowledge. Thus, the alternative hypothesis suggested a positive relationship between condition severity and youth's knowledge over time.

Method

Participants

Participants consisted of 70 families with an 8- to 9-year-old child with SB at Time 1 (T1; 39 males, 31 females; mean age = 8.33 years; 80% Caucasian), all of whom were part of a larger longitudinal study investigating family relationships and psychosocial adjustment in children and adolescents with SB (Holmbeck, Coakley, Hommeyer, Shapera, & Westhoven, 2002; Holmbeck, Johnson et al., 2002; Holmbeck and Weshoven, 2001; Holmbeck et al., 2003). In all families, biological mothers (mean age = 37.71 years) participated at T1, and in 56 families, fathers (mean age = 40.91 years) participated at T1. The sample included a range of family income levels (33%, <$40,000; 24%, $40,000–49,999; 39%, >$50,000; 4%, missing). Children's mean peabody picture vocabulary test score (PPVT-R; Dunn & Dunn, 1981) was 93.14 (*SD* = 18.66, range = 40–143).

Information concerning several physical status variables was collected from maternal report and medical chart reviews including: (a) spinal lesion level (medical chart): 32% sacral, 54% lumbosacral or lumbar, 13% thoracic (interrater reliability = 83%); (b) SB type (medical chart): 82% myelomeningocele, 12% lipomeningocele, 6% other (interrater reliability = 92%); (c) shunt status (maternal report): 71% shunt and 29% no shunt; (d) ambulation (maternal report): 43% use no assistance or ankle-foot orthoses (AFOs) for ambulation more than 50% of the time, 40% ambulated with knee-ankle-foot orthoses (KAFOs) or hip-knee-ankle-foot orthoses (HKAFOs) more than 50% of the time, and 17% used a wheelchair for ambulation more than 50% of the time; and (e) average number of shunt surgeries (other than original shunt placement surgery) among those with shunts (medical chart): mean = 2.50, *SD* = 2.91 (interrater reliability = 97%). [Interrater reliabilities for all variables obtained via medical chart review (i.e., spinal lesion level, SB type, and average number of shunt surgeries) are reported. For each of these variables, the percent agreement between raters was used as an index of interrater reliability. For shunt status and ambulation status, no interrater reliabilities are reported as these variables were obtained via maternal report on a background questionnaire.]

Participant Recruitment

Children with SB between the ages of 8 and 9 years were recruited from two children's hospitals within a large Midwestern city, a university-based medical center, and a statewide SB association (for detailed information about the recruitment strategy, see Holmbeck, Coakley et al., 2002). Of the 132 eligible families, 62 declined participation, resulting in a final sample of 70 families at T1. The relatively high decline rate was attributable, in part, to the extensive time commitment asked of participants (i.e., a longitudinal study consisting of several 3-hr family visits). Analyses revealed no significant differences between families who participated and families who declined to participate in terms of child lesion levels [$\chi^2(2) = .62$, $p > 0.05$] or type of SB (myelomeningocele versus lipomeningocele) [$\chi^2(1) = 1.63$, $p > 0.05$]. Of the 70 families who participated at T1, 67 were retained at T2 and 64 participated at T3.

Procedure

Data for this study represent the first three waves of data collection from a larger longitudinal study following children with SB through middle childhood and adolescence. Information was obtained during three separate 3-hr home visits conducted by trained research assistants, the first of which occurred when children were 8–9 years of age (T1), the second when children were 10–11 years old (Time 2, T2), and the third when children were 12–13 years old (Time 3, T3). Families were compensated for participation at each time point. Across all time points, a similar method of data collection was utilized whereby informed consent (or child assent) was obtained from both parents and child at the start of the visit. Then, questionnaire packets were completed by parents. Children also completed questionnaires that were read aloud to them using an interview format to enhance comprehension at T1 and as needed at subsequent

time points (most children at T2 and T3 completed questionnaires independently). Additionally, medical chart reviews were conducted at T1 to ascertain data about condition severity and associated medical complications.

Measures

Independent Variable: IQ

The PPVT-R (Dunn & Dunn, 1991) was used as a measure of children's verbal intelligence at T1. The PPVT-R is a receptive vocabulary task in which a word is presented verbally and the child is asked to choose the one picture that corresponds to the spoken word from four possibilities. For the purposes of the present investigation, the child's standard score was used.

Independent Variable: Condition Severity

A composite rating of condition severity was computed by assessing the following condition parameters: (a) lesion level based on the median of the three most recent reports in the medical chart (sacral, lumbar, or thoracic); (b) type of SB as indicated in the medical chart (myelomeningocele, lipomeningocele, or other); (c) shunt status as reported by mother; and (d) ambulation status as reported by mother (no assistance/ankle-foot orthotics, knee-ankle-foot orthotics/hip-knee-ankle-foot orthotics, or wheelchair). Severity scores ranged from 4 to 10 (mean = 7.01, *SD* = 1.77), with higher scores indicative of greater severity. Composite scores were computed using the following coding scheme: shunt status (no = 1, yes = 2), myelomeningocele type (no = 1, yes = 2), lesion level (sacral = 1, lumbar = 2, thoracic = 3), and ambulation status (no assistance/ankle-foot orthotics = 1, knee-ankle-foot orthotics/hip-knee-ankle-foot orthotics = 2, wheelchair = 3).

Dependent Variable: Condition Knowledge

Children's knowledge of SB was assessed with an 18-item questionnaire modeled after Wills et al. (1993). In the present investigation, 12 of the 18 items administered in the current study were used. Six items that did not address children's factual knowledge of SB and associated impairments were not included in the present analyses. The 12 chosen items addressed three domains of knowledge: (a) etiology of SB, (b) physical manifestations or functional impairments associated with SB (functional status), and (c) issues related to shunt functioning (see Table I for a list of all questions used). Children were asked to write their responses to a series of open-ended questions.

To evaluate the content of children's knowledge empirically, responses to each question were broadly categorized as correct (2 points), inconclusive (1 point; a response with some elements of a correct response but either somewhat vague or developmentally immature), or incorrect (0 points) based on a review of factual information about the clinical presentation of SB. Within the above broad categories, responses were further classified into content-based categories. The procedure of classifying responses by content category was performed for each of the 12 knowledge questions utilized in the current investigation, and Tables II–IV list the content-based subcategories by question. Coding of the content-based subcategories was performed as follows. The first three authors collaboratively generated content domains, which were then independently rated by a trained graduate research assistant. Analyses of rater reliability for the assignment of responses to the content-based subcategories suggested high levels of agreement (kappa mean = .79, range = .61-.96) on all items.

Results

Plan of Analysis

To test the hypothesis of significant gains in accuracy of knowledge over time, repeated measures ANOVAs were conducted to compare scores across T1, T2, and T3 for each item, and paired *t* tests were used to probe significant multivariate findings. For questions related to etiology and functional impairment, analyses were conducted with the full sample of children. In contrast, for items related to shunt functioning, the sample was restricted to children with shunts (n = 48), because children without shunted hydrocephalus were not expected to possess knowledge in this domain. To document the content of children's responses over time, follow-up analyses were conducted to assess the frequency of each category of response for each item across T1, T2, and T3.

To examine predictors of knowledge change, hierarchical regression analyses were conducted whereby T2 composite knowledge was entered into a first block (in an effort to control for previous level of knowledge and to allow for an examination of knowledge change), followed by the independent variable of interest (either IQ or condition severity) in a second block, with T3 composite knowledge as the outcome variable. Composite knowledge ratings for T2 and T3 were calculated by summing the number of correct and inconclusive responses across all items for each child, where higher scores reflected more knowledge. This composite variable evidenced adequate internal consistency (T2 α = .77, T3 α = .65).

Accuracy of Knowledge Over Time

A series of repeated measures ANOVAs were conducted to assess changes in the accuracy of children's knowledge over time. Table I provides mean scores for children on each item across all three time points. Findings revealed that on 6 of 12 items, the accuracy of children's knowledge improved significantly from T1 to T3. In the etiology domain, gains were made across T1 to T3 in children's understanding of the age at which SB develops ["How old do you have to be to get SB?" $F(2,59) = 6.90$, $p = 0.00$], the process by which SB is acquired ["How do kids get SB?" $F(1,60) = 4.21$, $p = 0.05$], and the possibility of offspring having SB ["If someone with SB has a child, will the child have SB too?" $F(2,59) = 3.21$, $p = 0.05$]. In contrast, in the etiology domain, no evidence for gains in knowledge was documented in children's understanding of whether or not SB can develop in adulthood ["Can people get SB as adults?" $F(2,59) = 2.29$, $p = 0.11$].

In the functional status domain, significant gains, in terms of accuracy of knowledge, were made in children's understanding of the possible impairments in bladder functioning ["How do kids with SB usually have to go to the bathroom?" $F(2,59) = 5.18$, $p = 0.01$] and ambulation ["Why cannot kids with SB walk quite right?" $F(2,59) = 5.15$, $p = 0.01$] associated with SB. However, no gains in knowledge were documented in children's understanding of characteristics that differentiate children with SB from able-bodied children ["What makes kids with SB different from other kids?" $F(2,59) = 1.526$, $p = 0.23$] or in children's understanding of the consequences of forgetting to catheterize ["What happens if they forget to catheterize?" $F(2,59) = 0.98$, $p = 0.38$].

Finally, among children with shunts, gains were evident in understanding the function of a shunt from T1 to T3 ["What does a shunt do?" $F(2,40) = 5.18$, $p = 0.01$]. No significant changes in children's understanding of (a) signs of shunt malfunction ["How does it feel if a shunt is not working right?" $F(2,40) = 0.59$, $p = 0.56$], (b) location of a shunt ["Where is a shunt located?" $F(2,40) = 0.93$, $p = 0.41$], or (c) medical management of shunt malfunction ["What do doctors do if a child's shunt is not working right?" $F(2,40) = 0.75$, $p = 0.48$] were documented. With respect to the last two questions, the lack of improvement in accuracy of knowledge was likely because of children showing high levels of understanding across all three time points.

Content of Children's Knowledge Over Time

To determine the content of children's responses over time, frequency analyses were conducted (see Tables II–IV). As indicated in Table I, within the etiology domain, children's understanding of how old one must be to get SB increased significantly across time points, as did their understanding of how SB develops, and whether offspring of individuals with SB will necessarily develop the condition. Despite these gains, however, an analysis of the content of children's responses at each time point revealed significant gaps in knowledge, even at ages 12–13 (T3; see Table II). Specifically, only 50% of responses at T3 demonstrated an understanding that SB is contracted

Table I. Group Means (*SD*) for Repeated Measures ANOVAs[a]

Question	Time 1	Time 2	Time 3	ANOVA
Etiology				
How old do you have to be to get SB?	.49 (.87)	.85 (.99)	1.05 (1.01)	T2>T1*, T3>T1**
Can people get SB as adults?	.72 (.97)	.59 (.92)	.82 (.99)	*ns*
How do kids get SB?	00 (.00)	.00 (.00)	.13 (.50)	T3>T2*, T3>T1*
If someone with SB has a child, will the child have SB too?	1.11 (1.00)	1.05 (1.01)	1.44 (.90)	T3>T1*, T3>T2*
Functional differences				
What makes kids with SB different from other kids?	1.31 (.96)	1.02 (1.01)	1.18 (.99)	*ns*
How do kids with SB usually have to go to the bathroom?	1.08 (1.01)	1.41 (.92)	1.54 (.85)	T2>T1*, T3>T1**
What happens if they forget to catheterize?	.85 (1.00)	.95 (1.01)	1.08 (1.00)	*ns*
Why cannot kids with SB walk quite right?	.39 (.80)	.59 (.92)	.82 (.99)	T3>T1**
Shunt functioning				
Where is a shunt located?	1.76 (.66)	1.86 (.52)	1.90 (.43)	*ns*
What does a shunt do?	.48 (.86)	.43 (.83)	1.00 (1.01)	T3>T1*, T3>T2**
How does it feel if a shunt is not working right?	.71 (.97)	.71 (.97)	.90 (1.01)	*ns*
What do doctors do if a child's shunt is not working right?	1.52 (.86)	1.71 (.71)	1.71 (.71)	*ns*

ns, nonsignficant; SB, spina bifida.

[a]Scores ranged from 0 to 2. 0 = incorrect, 1 = inconclusive, 2 = correct.

*$p < .05$.

**$p < .01$.

Table II. Etiology: Content of Adolescent Responses and Frequencies[a]

Etiology questions	Time 1 ($n = 70$)	Time 2 ($n = 67$)	Time 3 ($n = 64$)
How old do you have to be to get SB?			
Correct			
SB is contracted prenatally	25%	41.2%	50.8%
Incorrect			
SB can be contracted at any age	32.8%	22.1%	24.6%
SB is contracted during infancy (ages from birth–2 years)	12.5%	5.9%	6.2%
SB is contracted during childhood (ages 3–12 years)	9.4%	2.9%	3.1%
SB is contracted during adolescence/adulthood (age >13 years)	3.1%	0.0%	0.0%
Do not know/other	12.5%	27.9%	13.8%
Can people get SB when they are adults?			
Correct			
No	35.9%	30.9%	40.0%
Incorrect			
Yes	48.4%	36.8%	30.8%
Uncertain	1.6%	8.8%	12.3%
Do not know/other	12.5%	22.1%	13.8%
How do kids get SB?			
Correct			
Inherited, genetic	0%	0%	1.5%
Folic acid deficiency	0%	0%	1.5%
Cause is unknown	0%	1.5%	3.1%
Inconclusive			
Birth defect, vague	0%	1.5%	3.1%
Incorrect			
Birth complication	1.6%	2.9%	1.5%
Postnatal illness/injury	3.1%	1.5%	0%
Back problems	7.8%	5.9%	6.2%
Do not know/other	89.1%	80.9%	78.5%
If someone with SB has a child, will the child have SB too?			
Correct			
No	32.8%	33.8%	23.1%
Possibly	23.4%	19.1%	47.7%
Inconclusive			
Vague response	3.1%	1.5%	3.1%
Incorrect			
Yes	20.3%	5.9%	1.5%
Do not know/other	18.8%	39.7%	23.1%

SB, spina bifida.

[a]Frequencies do not necessarily add to 100% because some youth in this study provided more than one response per item. A maximum of three responses were coded for each child.

prenatally. Common misperceptions at T3 regarding the etiology of SB included the belief that it could be contracted at any age (25% of responses) or that individuals could develop SB as adults (31%). Moreover, by T3, only 6% of responses indicated that the cause of SB was (a) inherited or genetic, (b) associated with a folic acid deficiency, or (c) that the specific etiology was largely unknown. Finally, 23% of 12- to 13-year-old responses indicated uncertainty about whether or not someone with SB will inevitably conceive a child who also has SB.

Table III summarizes the content of children's responses concerning functional status. Across T1 to T3, children evidenced a stable, moderate level of knowledge about ways in which children with SB differ from able-bodied children (i.e., "What makes kids with SB different from other kids?"). Across all time points, children tended to describe physical differences that are apparent to others or medical/biological differences more frequently than they described differences in self-help skills, cognitive functioning, or social functioning. Regarding

Table III. Functional Status: Content of Adolescent Responses and Frequencies[a]

Functional status questions	Time 1 ($n = 70$)	Time 2 ($n = 67$)	Time 3 ($n = 64$)
What makes kids with SB different from other kids?			
Correct			
Specific physical difference apparent to others	46.9%	41.2%	38.5%
Medical/biological difference (not apparent to others)	12.5%	11.8%	16.9%
Difference in self-help skills	3.1%	1.5%	1.5%
Cognitive difference (school, learning, and attention)	0%	0%	1.5%
Social difference	4.7%	0%	0%
Inconclusive			
Vague response	12.5%	19.1%	29.2%
Incorrect			
No difference exists	3.1%	7.4%	4.6%
Have abilities that other children do not have	1.6%	1.5%	0%
Do not know/other	23.4%	22.1%	15.4%
How do kids with SB usually have to go to the bathroom?			
Correct			
Use of toileting program/device	48.4%	55.9%	67.7%
Requires assistance from others	40.6%	52.9%	67.7%
Toileting done in same manner as kids without SB	1.6%	8.8%	4.6%
Inconclusive			
Indication of variability	0%	0%	1.5%
Indicates difference, but vague (e.g., not on toilet)	9.4%	8.8%	0%
Incorrect			
Indicates time of day or how frequently	1.6%	2.9%	1.5%
Do not know/other	28.1%	20.6%	15.4%
What happens if they forget to catheterize?			
Correct			
Physical illness or infection	21.9%	16.2%	18.5%
Bladder/bowel incontinence	18.8%	27.9%	30.8%
Physical discomfort	6.3%	7.4%	4.6%
Urine retention	1.6%	0%	3.1%
Inconclusive			
Nonspecific negative outcome (e.g., something bad)	3.1%	2.9%	3.1%
Child will be punished	3.1%	2.9%	0%
Incorrect			
Do not know/other	43.8%	32.4%	30.8%
Why cannot kids with SB walk quite right?			
Correct			
Reduced sensation/muscle tone in feet or legs	14.1%	4.4%	7.7%
Problems with CNS	9.4%	23.5%	30.8%
Inconclusive			
Indication of back, feet or legs, but answer is vague	20.3%	22.1%	7.7%
Require appliance/brace for walking	12.5%	0%	3.1%
Walking problems are part of having SB	12.5%	1.5%	4.6%
Other (e.g., surgery)	4.7%	2.9%	1.5%
Incorrect			
There are no problems with walking	4.7%	1.5%	1.5%
Specific, but wrong response (e.g., leg is broken)	4.7%	5.9%	0%
Do not know/other	29.7%	36.8%	36.9%

CNS, central nervous system; SB, spina bifida.

[a]Frequencies do not necessarily add to 100% because some youth in this study provided more than one response per item. A maximum of three responses were coded per item for each child.

Table IV. Shunt Functioning: Content of Adolescent Responses and Frequencies[a]

Shunt functioning questions	Time 1 ($n = 44$)	Time 2 ($n = 48$)	Time 3 ($n = 46$)
Where is a shunt located?			
Correct			
Head	79.5%	75%	84.8%
Head extending to stomach	6.8%	14.6%	6.5%
Inconclusive			
Vague response	2.3%	6.3%	2.2%
Incorrect			
Do not know/other	9.1%	4.2%	4.3%
Drains and (includes one detail about function)	12.5%	14.7%	27.7%
What does a shunt do?			
Correct			
Drains and (includes one detail about function)	12.5%	14.7%	27.7%
Helps brain to work better/decreases symptoms	4.7%	1.5%	4.6%
Pumps fluid/drains	1.6%	0%	6.2%
Inconclusive			
Nonspecific positive impact (e.g., prevents getting sick)	6.3%	5.9%	3.1%
Incorrect			
Nonspecific medical purpose (e.g., brings blood to head)	3.1%	8.8%	4.6%
Causes pain/creates problems	1.6%	1.5%	3.1%
Do not know/other	60.9%	47.1%	38.5%
How does it feel if a shunt is not working right?			
Correct			
Specific symptoms (e.g., dizzy and headache)	26.6%	26.5%	30.8%
Inconclusive			
Generalized pain or illness	18.8%	16.2%	12.3%
Need for intervention	1.6%	2.9%	1.5%
Other (e.g., death)	4.7%	4.4%	3.1%
Incorrect			
Do not know/other	45.3%	38.2%	44.6%
What do doctors do if a child's shunt is not working right?			
Correct			
Requires a test, procedure, or to be checked	43.2%	47.9%	47.8%
Needs to be fixed or replaced	40.9%	35.1%	39.1%
Inconclusive			
Need for intervention, but vague (e.g., put in an IV)	6.8%	8.3%	4.3%
Incorrect			
Do not know/other	13.6%	8.3%	10.9%

[a]Questions include only participants with shunts. Frequencies do not necessarily add to 100% because some youth in this study provided more than one response per item. A maximum of three responses were coded per item for each child.

catheterization, by T3, 68% of responses to a question assessing knowledge of the process of bowel/bladder voiding (i.e., "How do kids with SB usually have to go to the bathroom?") indicated an understanding that a toileting program or device (e.g., catheter) must be utilized by some individuals with SB, as well as an understanding that children with SB typically require assistance from others in bladder and bowel management tasks. Moreover, children evidenced a stable, moderate level of knowledge about the consequences of inconsistent catheterization over time (i.e., "What happens if they forget to catheterize?"), and by T3, about 55% of responses correctly indicated at least one negative consequence of missed catheterization. However, over 30% of responses at T3 were incorrect, suggesting a lack of understanding of negative consequences of missed catheterization. Additionally, over time, when asked about the ambulation problems often associated with SB (i.e., "Why cannot kids with SB walk quite right?"), children's responses suggested improved understanding that difficulties in ambulation are secondary to central nervous system (CNS) impairment or to orthopedic/muscle tone issues

from T1 to T3. Although some gains were evident, 38% of children's responses at T3 indicated that they did not know or suggested an incorrect reason for why children with SB often have difficulty with ambulation.

Children's understanding of shunt functioning was evaluated only among those children with shunts ($n = 48$). As indicated in Table IV, these children demonstrated consistently high levels of knowledge about shunt location across all three time points. Specifically, 91% of responses at T3 indicated that a shunt was located in the child's head, whereas 7% of responses evidenced a more advanced understanding that it extends from the head to the stomach. In contrast, children's understanding of what a shunt does and of warning signs of shunt malfunction was less well developed. Although children made gains in their understanding of the function of a shunt across T1 to T3, only 39% of responses were fully correct definitions (e.g., drains fluid, decreases symptoms, and helps brain to work better) by T3. Additionally, children showed consistently poor understanding of the warning signs of shunt malfunction over time. In fact, by T3, only 31% of responses articulated a specific warning sign of shunt malfunction. Finally, children demonstrated consistently high levels of understanding of a doctor's role in treating shunt malfunction across all three time points. In fact, 87% of responses at T3 correctly indicated that a specific form of medical intervention was necessary to manage shunt malfunction.

Predictors of Knowledge

A series of multiple regression analyses were conducted to evaluate predictors of changes in children's overall condition-related knowledge from T1 to T3. Overall, analyses failed to offer support for the hypotheses that either IQ level [F *change*$(1,58) = .026$, $p = 0.872$] or condition severity [F *change*$(1,57) = .093$, $p = 0.762$] was positively associated with T3 knowledge above the variance accounted for by T2 knowledge. Similarly, bivariate correlations between IQ and knowledge at T1 ($r = 0.073$, $p = 0.569$), T2 ($r = –0.071$, $p = 0.567$), and T3 ($r = –0.043$, $p = 0.734$) failed to support a significant relationship between cognitive functioning and knowledge. Bivariate correlations between severity and T1 ($r = 0.077$, $p = 0.549$), T2 ($r = –0.077$, $p = .540$), and T3 knowledge ($r = 0.083$, $p = 0.517$) were also nonsignificant.

Discussion

The primary purpose of this longitudinal study was to evaluate changes in accuracy of condition-related knowledge over time among preadolescents with SB. Specifically, this study examined knowledge changes within the content areas of etiology, functional status, and shunt functioning. Additionally, a detailed content analysis was conducted to provide information regarding the quality of child responses at each of three time points. Finally, the utility of child IQ and condition severity in predicting change in knowledge was evaluated.

In general, most condition domains were characterized by low-to-moderate levels of knowledge accuracy across time. Although youth in this sample demonstrated significant gains in knowledge over time in some areas, the quality and accuracy of condition-related knowledge demonstrated in this sample indicated that there was a lack of age-appropriate understanding of fundamental information across condition domains. An examination of the accrued knowledge over time indicates that youth made the most significant gains in knowledge related to the domains of etiology and functional status. In general, children were most successful at gaining knowledge in the area of etiology, with results indicating that as youth got older, they gained a better understanding of the origin and transmission of SB. This finding supports past research, demonstrating that adolescents are better able to understand concepts of causation and disease transmission (Berry et al., 1993; Brewster, 1982; La Greca et al., 1990; Perrin & Gerrity, 1981). Though results indicated that children made significant gains in this domain, preadolescents at T3 continued to have a much lower than expected accuracy regarding knowledge of the etiology of their condition. For example, at T3, fewer than 10% of responses provided fully correct information regarding how SB is contracted. Within the functional status domain, a similar pattern emerged. Gains in knowledge were evident in youth's understanding of why children with SB walk differently from their able-bodied peers and in regard to their toileting behavior. Though responses indicated a statistically significant increase in accuracy from T1 to T3, many responses included incorrect or vague information regarding ambulation differences. In the area of shunt functioning, youth demonstrated gains in their basic understanding of the purpose of a shunt. However, as in other areas of demonstrable gain, most responses indicated an incomplete understanding of shunt functioning. Thus, although there was evidence that gains in knowledge accuracy were made over time, the content analysis provided clinical data indicating that young adolescents with SB continued to have incomplete or incorrect information regarding the etiology, functional status (e.g., catheterization), and shunt functioning related to their condition.

In summary, findings regarding changes in knowledge over time indicate that increases in age were associated with more accurate knowledge for half of the questions polled on our questionnaire. Despite making statistically significant gains, it is concerning that there remains a paucity of condition-related knowledge across domains. These findings support past research with other pediatric groups, demonstrating that deficits in children's understanding of their medical condition are common (Celando et al., 1998; Ievers-Landis & Drotar, 2000).

Several items that did not evidence significant gains in knowledge represented areas of knowledge that remained consistently accurate across time. The lack of significant gains in knowledge for these items reflects that most youth understood the concepts at an early age and maintained their knowledge over time. Specifically, the majority of responses correctly indicated the location of a shunt, what doctors do if a shunt is not working correctly, and what makes kids with SB different from other kids. On the other hand, there were three questions in this study with responses that consistently reflected poor knowledge across time. Specifically, the vast majority of youth in this sample did not have accurate information regarding items that relate to medical functioning, illness transmission, and consequences of missed catheterization (i.e., "How does it feel if a shunt is not working" right?; "Can people get SB when they are an adult?;" and "What happens if they forget to catheterize?").

In contrast to past research, this study did not find a relationship between IQ and changes in knowledge. Research in this area of pediatric psychology has typically found that cognitive status is a better predictor of knowledge than is age (Berry et al., 1993; Feldman & Varni, 1985; Perrin & Gerrity, 1981). It is possible that our failure to replicate this finding indicates a weakness in our assessment of IQ. In this study, IQ was assessed via PPVT scores obtained at T1 data collection. While PPVT scores are typically highly correlated with verbal IQ, future investigations may be strengthened by the inclusion of a more comprehensive cognitive assessment. This study also failed to support a relationship between condition severity and knowledge. It is possible that the small sample size limited our ability to detect this relationship. It is also possible that we lacked sufficient variability in condition severity to predict differences related to gains in knowledge. Specifically, 64% of participating children had severity ratings of 6, 7, or 8 (range 4–10), suggesting that our sample was overrepresented by individuals of moderately high condition severity. Future research may help to clarify whether severity emerges as a significant predictor of knowledge in samples of youth with greater variability in condition severity.

This study has certain limitations that have implications for future research. First, although the study employed a longitudinal design, we studied a relatively narrow age range of participants. Therefore, it is possible that gains in knowledge would be more evident in follow-up studies with this sample. Second, the generalizability of these findings is limited primarily to Caucasian, middle-class families. Third, the questions in our knowledge questionnaire assessed factual condition-related knowledge. It would be useful to also assess pragmatic condition-related knowledge (e.g., how to perform intermittent self catheterization) to ascertain how adept youth are at understanding and performing the day-to-day care that is required to maintain their good health. Moreover, to a certain extent, children's responses may have been dependent upon the wording and/or format of the question (e.g., at T3, 31% of children reported that someone can get SB as an adult in response to question that could be satisfied with a "yes" or "no" response; however 0% of participants at T3 spontaneously offered a response that SB could be contracted during adolescence or adulthood in response to the open-ended question that asked about how old a person must be to develop SB). Thus, using questions that elicit open-ended responses as a means of assessing knowledge may reduce the likelihood that children will guess and may be a preferred method of assessment. One potential drawback to the use of open-ended questions, however, is that the quality of the response may be influenced by a child's writing ability. As such, a flexible administration method whereby children can either write their own responses or have the responses transcribed for them may be a preferred assessment strategy.

The clinical implications of this study have relevance to health care providers and parents of youth with SB. Findings from this study indicate that a majority of youth with SB have significant gaps in knowledge regarding the domains of etiology, functional status, and shunt functioning. Given the body of past research suggesting that children's knowledge of their own condition may positively impact coping skills, reduce anxiety, increase feelings of self-efficacy, and facilitate medical adherence (Bartholomew et al., 1991; Edwinson et al., 1988; Lorenz et al., 1985), it is important that youth with SB have ongoing education (ideally in the context of a routine medical appointment) to ensure exposure to accurate and age-appropriate condition-related knowledge. In a congenital condition like SB, it is possible that health care providers and parents overlook the need to

provide ongoing comprehensive education. Instead, there may be more attention to education for chronic illnesses that develop in middle childhood or adolescence (e.g., Crohn's disease, cancer and rheumatoid arthritis). In other words, a new diagnosis may prompt an educational intervention, whereas a congenital condition may not. Underscoring the need to provide multiple developmentally appropriate educational sessions to youth with SB to health care professional colleagues may be an important role of pediatric psychologists in applied medical settings. Greater attention to parent education could help to increase youth understanding of etiological factors, functional strengths and weaknesses, and issues related to medical care. Additional education about catheterization and shunt malfunction are two domains that may be of particular clinical significance. This type of ongoing education may be especially relevant for adolescents, as it will help to foster competence as they transition toward assuming increased responsibility for their health care and well-being.

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