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Condition-related knowledge among children and adolescents with spina bifida in a Swedish county

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Spina bifida is a congenital birth defect, resulting in physical and cognitive dysfunctions. Condition-related knowledge among children and adolescents with spina bifida is essential to facilitate independent management of their condition. The aim was to describe the condition-related knowledge among children and adolescents with spina bifida in a Swedish county. Thirteen persons with spina bifida (10 to 17 years) participated. Condition-related knowledge was assessed (n = 13) using a questionnaire (KOSB) and a semi-structured interview (n = 8). Interview data were analyzed using qualitative content analysis. The participants had well-developed knowledge concerning proper bladder management, but were lacking knowledge of signs of shunt malfunctioning and etiology. Some participants were uninterested in learning about their condition, despite being aware that they lacked knowledge. The findings indicate potential areas that may be included in local educational initiatives. It should be considered that persons with spina bifida may not be motivated to learn more about their condition.

Keywords: spina bifida; children; adolescents; condition knowledge; qualitative content analysis

Introduction

Spina bifida is a congenital birth defect originating from an incomplete closure of the embryonic neural tube during the first month of foetal development. Despite the practice of surgically closing the tube upon birth, neurological injuries to the spinal cord are unavoidable (Cavalheiro et al. 2008). Spina bifida results in varying levels of motor and cognitive impairments, as well as neurogenic bladder and bowel dysfunction (Dennis and Barnes 2010; Olsson et al. 2007). In most cases a subsequent development of hydrocephalus occurs (Olsson et al. 2007), which requires a shunt to drain the excess cerebrospinal fluid from the ventricles. The causative mechanism of spina bifida has not yet been established and various genetic and environmental factors (such as lack of folic acid) have been suggested to be pathogenetic (Blom et al. 2006; Cabrera et al. 2004). The most severe form of spina bifida is myelomeningocele (MMC) (Josan, Morokoff, and Maixner 2008).

As individuals with spina bifida pass through the transition from being children to becoming independent adults, they seem to have problems with attaining skills of

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autonomy (Davis et al. 2006; Friedman et al. 2009). One important factor in this process is obtaining knowledge about their functional impairment, which may have a positive effect on coping with the disability. Condition-related knowledge among children and adolescents with various diagnoses, such as cystic fibrosis and diabetes, has been documented to have a positive effect on their psychosocial adjustment. Distress and anxiety decrease and the child's self-management and adherence to the medical regimen increase (Edwinson, Arnbjörnsson, and Ekman 1988; Lorenz, Christensen, and Pichert 1985; Stapleton et al. 2000).

Condition-related knowledge among people with spina bifida has been studied previously. Dunn, McCartan and Fuqua (1988) found that young children with spina bifida and other disabilities had started to develop an understanding of their disability, and suggest that preschool children may be ready for basic level information. Erickson (1992) reported that adolescents and young adults with spina bifida lacked knowledge about fundamental aspects and were relatively uninformed about precautionary measures. Feldman and Varni (1985) found that children with spina bifida knew more about spina bifida than about health and illness in general. In a longitudinal study of children with spina bifida Greenley et al. (2006) showed that the children's condition-related knowledge increased over time, but many participants still lacked an age-appropriate understanding. A majority of the participants partly lacked knowledge about etiology, functional status and shunt functioning. Two clinically relevant areas, namely catheterization and shunt malfunction, were found to be in particular need of additional education. As suggested by Johnson (1984), knowledge in one condition-specific content area is not necessarily predictive of knowledge in another area. This notion emphasizes the need to assess condition-related knowledge across multiple domains.

Some studies have been conducted concerning the experiences of Swedish children and adolescents with spina bifida (e.g. Fägerskiöld and Glad Mattsson 2010; Skär 2003). However, a literature search did not reveal any published studies on condition-related knowledge in a Swedish context. Investigating condition-related knowledge could yield important implications for efforts to understand and improve independence among young people with spina bifida. Therefore the aim of the present study was to describe the condition-related knowledge among children and adolescents with spina bifida in a Swedish county.

Methods

The study presented here is based on data collected in a project that investigated condition-related knowledge and emotional and social adjustment among children and adolescents with spina bifida in a Swedish county. A mixed methods approach was taken, collecting data through questionnaires and interviews to allow a triangulation of obtained results from different sources (Creswell 2009). Two questionnaires were employed in the project, namely: Knowledge of spina bifida questionnaire (KOSB) (Greenley et al. 2006), and Beck Youth Inventories (Beck, Beck, and Jolly 2004). The latter questionnaire is used to assess emotional and social impairments in children and adolescents. The interview questions addressed three main areas: participants' 1) condition-related knowledge, 2) experiences of living with spina bifida, and 3) ideas about future life. The present study describes results from KOSB and the condition-related knowledge content area of the interviews.

Participant recruitment

The data were collected from 2008 to 2009. Children and adolescents aged 10 to 18 years with spina bifida (with or without hydrocephalus) from a county in Sweden were recruited for the study. To ensure that participants would be able to understand the questions asked, the criteria for enrolment included a lower age limit of ten years and that participants should not have intellectual disabilities more severe than the diagnosis mild intellectual disability (i.e. ICD-10 code F70).

Participants were recruited from a clinic where individuals with spina bifida are referred to for monitoring and managing bowel and bladder functions. All individuals with spina bifida in the target county are referred to this clinic, and it was thus expected that the entire population of children and adolescents with spina bifida that fulfilled the inclusion criteria were included in the recruitment process. Information about the study and a request to participate was sent to all the children and adolescents (and their caregivers), that fulfilled the inclusion criteria, and who resided in the target county (n = 15). Two eligible participants declined participation at this stage; one because of a stressful situation, and one because the person was not interested in taking part in the study. Thus 13 persons and their caregivers consented to participate in the study. Eight of these participants were asked to take part in an interview in addition to filling out a questionnaire. After performing eight interviews, it was considered that the collected interview material was sufficiently rich for the purpose of the present study. Therefore, the remaining five only completed a questionnaire. Background information about each participant was collected from his or her respective caregiver (Table 1). None of the participants were reported to have any intellectual disability diagnosis. The term spina bifida is used in this report to refer to the disability of the participants, all of whom had MMC.

The regional research ethical committee of Linköping approved the study on 17–06–2005 (M67-05).

Table 1. Background information about the children and adolescents with spina bifida (n = 13) that participated in the study. The age range of the participants was 10–17 years, with mean and median ages of 14. The same age parameters characterized the interviewed subset (n = 8).

Background variables	Questionnaire (n = 13)	Interview (n = 8)
Gender		
Female	6	4
Male	7	4
Shunt		
Yes	11	6
No	2	2
Use of clean intermittent catheterization		
Yes	12	7
No	1	1
Ambulatory status		
walk without aid	4	4
walk with aid or unable to walk (e.g. wheelchair or other assistive device)	9	4

Data collection

Knowledge of spina bifida questionnaire

The following three knowledge domains are addressed by KOSB (Greenley et al. 2006; O'Mahar et al. 2010): (a) etiology, (b) functional status (physical manifestations including functional impairments), and (c) specific issues concerning shunt function. It contains open-ended questions to which participants provide written responses. Two of the twelve items in Greenley et al. (2006) (both related to the etiology domain) were omitted, namely *'How old do you have to be to get spina bifida?'* and *'If someone with spina bifida has a child, will the child have spina bifida too?'*. The reason for this omission was concerns that the two items after translation into Swedish would bias the respondents' answers. The subject of etiology was covered by two of the remaining ten items. To ensure that an accurate translation of the items was made, a procedure that consisted of independent translation and back-translation was used (Streiner and Norman 2003). The back-translated items were similar or identical to the original wording.

All 13 participants completed KOSB, most of them (n = 8) with a researcher present before taking part in the interview. The questionnaires were administered by mail to the other participants (n = 5) together with instructions emphasizing that the child should complete it without assistance.

Interviews

A semi-structured interview was conducted (Kvale 2009), using an interview guide with a set of main questions that the interviewer followed up with additional questions to elicit more detailed responses. Questions were rephrased when necessary to ensure that the interviewees comprehended the questions as intended. The decision to perform semi-structured interviews was based on the presumed language skills of the study group. Children with spina bifida often have a specific language skill profile, involving deficits in expressive discourse (Dennis, Jacennik, and Barnes 1994), and a conversational style that has been described as containing irrelevant information and being referentially underspecified (Dennis et al. 2006). The semi-structured interview format was employed to help the interviewees to keep on topic and guide them through the different areas of the interview.

Depending on the preferences of the interviewees, two of the interviews were conducted in the home of the participant, while the remaining six interviews were conducted at a local health care facility that they were familiar with. Each interview lasted between 22–77 minutes (median 35 minutes), and were audio recorded. All interviews were conducted with only the interviewer (first author) and participant present. Distractions were kept at a minimum by conducting the interviews in undisturbed settings, and ensuring that adequate time was available for the interview.

Data analysis

Knowledge of spina bifida questionnaire

The responses on KOSB were categorized as correct, incorrect or inconclusive using the categories described by Greenley et al. (2006).

Interviews

The recorded interviews were transcribed verbatim and analyzed using qualitative content analysis (Graneheim and Lundman 2004). The transcribed interviews were read through several times to get a sense of the material. The parts of the interview transcripts that dealt with the participants' condition-related knowledge were identified and brought together into a separate text, which constituted the unit of analysis for this study. The text was divided into meaning units and condensed, taking the context into consideration. The meaning units consisted of single statements (parts or whole), or a short sequence of statements, which contained aspects that were related to each other by their context and content. The condensed meaning units were then labelled with appropriate codes, and the codes were sorted into sub-categories, categories and themes. This process consisted of several cycles of analysis and discussion between two of the researchers (author 1 and author 4) until agreement was achieved on how to label the codes, sort them into categories, and formulate themes. The second and third authors (author 2 and author 3) contributed to the analysis by checking the coding and categorization to obtain agreement within the research group on the way data were labelled and sorted. Because the study object was participants' expressed knowledge, the analysis primarily focused on the manifest content that can be discerned with relatively little interpretation.

Results

Knowledge of spina bifida questionnaire

All 13 participants responded to KOSB (Table 2). In the etiology domain the majority were unable to provide correct answers to the question '*How do kids get spina bifida?*' (Table 2). Some respondents gave an incorrect answer, for example by attributing the cause of spina bifida to chromosomal defects. In the functional status domain, the question '*Why cannot kids with spina bifida walk quite right?*' yielded many inconclusive answers (Table 2). Respondents referred to problems with the back, for example that the spinal column is broken or unstable. In response to the question '*What makes kids with spina bifida different from other kids?*' most correct responses referred to differences in physical functioning and toilet activities. However, three correct responses included differences in cognitive function, such as memory problems. In the shunt functioning domain, the majority of the responses to the questions '*What does a shunt do?*' and '*How does it feel if a shunt is not working right?*' were incorrect or inconclusive (Table 2). For the latter question, four of the respondents that answered incorrectly specified that they did not know any signs of shunt malfunctioning because they had never experienced any problems with the shunt.

Interview

Two main themes were found through the qualitative content analysis of the interviews: *Content of condition-related knowledge* (Table 3), concerning their practical and theoretical knowledge of spina bifida, and *Perceptions of condition-related knowledge* (Table 4), describing the participants' reflections about their own knowledge. The results are presented by theme, describing categories and sub-categories together with illustrative statements.

Table 2. Results from children and adolescents with spina bifida (n = 13) on the Knowledge of Spina Bifida questionnaire (KOSB) concerning condition-related knowledge. Participants that did not use clean intermittent catheterization or did not have a shunt were not asked all questions.

		Answer n = 13			
Domain	Question	Correct	Inconclusive	Incorrect	Not asked
Etiology	1. How do kids get spina bifida?	2	2	9	0
	2. Can people get spina bifida as adults?	7	1	5	0
Functional status	3. What makes kids with spina bifida different from other kids?	11	0	2	0
	4. How do kids with spina bifida usually have to go to the bathroom?	12	0	0	1
	5. What happens if they forget to catheterize?	11	0	1	1
	6. Why cannot kids with spina bifida walk quite right?	6	5	2	0
Shunt functioning	7. Where is a shunt located?	11	0	0	2
	8. What does a shunt do?	5	2	4	2
	9. How does it feel if a shunt is not working right?	5	1	5	2
	10. What do doctors do if a child's shunt is not working right?	10	0	1	2

Table 3. Categories and sub-categories for the theme 'Content of condition-related knowledge'.

Categories	Subcategories
Medical terms	Label of disability
	Shunted hydrocephalus
Etiology	Causal explanation
	Time of contraction
Physical effects	Movement
	Bowel and bladder
	Bodily maturation
	Sensory perception
Cognitive effects	Mathematical ability
	Executive functions
	Memory
Maintaining health	Toilet activities
	Physical exercise
	Skin sore management
	Achievement of independence
	No specific activities

Content of condition-related knowledge

Medical terms

The category "medical terms" is described in two subcategories. The subcategory label of disability describes how participants used the Swedish term for spina bifida (*ryggmärgsbråck*) to designate their disability, although not necessarily in conversations with others. Spina bifida also seemed to be used inappropriately as a label for conditions that imply movement disability but having other underlying causes. Participants with shunted hydrocephalus were aware that they have a shunt and some of them could explain why they have it. This subcategory contains descriptions of having no or very vague knowledge about hydrocephalus. It also contains explanations of what hydrocephalus is and its connection to a shunt:

> "It [hydrocephalus] is 'water on the brain' is it not? Well, it is when some fluid from the spine goes up to the head and stays up there, I do not really know why (...) It is the shunt that regulates that."

Table 4. Categories and sub-categories for the theme 'Perceptions of condition-related knowledge'.

Categories	Sub-categories
Attitude to own knowledge	Awareness of disability
	Assessment of knowledge state
	Opinion on information acquisition
Process of knowledge construction	Initial information
	Providers of further information
	Self-initiated information acquisition

Etiology

The category "etiology" is described in two subcategories. The subcategory causal explanation describes the origin of the condition as a congenital injury to the spine or back. Some participants mentioned nerves in their explanations of the condition:

> "There is something wrong with my back. It is something with the nerves in the back (inaudible) that is not as it should be."

The time of contraction subcategory contains descriptions of the condition as being contracted either before or during birth. Although many knew that the condition was congenital in their individual cases, some may not have generalized this to all cases of spina bifida. Awareness of being born with "damage to the spine" was expressed, while at the same time attributing the movement impairment to a surgical procedure several years later. Furthermore, a view of the condition as congenital was combined with claiming personal knowledge of an individual who had contracted spina bifida as an adult:

> "Mmm, but he could walk, before (. . .) But then it just went 'bang', and he was in a wheelchair."

Physical effects

The category "physical effects" is described in four subcategories. The subcategory movement describes how spina bifida gives rise to walking difficulties or the need for a wheelchair:

> "Mmm (pause) it is those [people with spina bifida] who do not, kind of, who cannot walk like others."

In the subcategory bowel and bladder, aspects such as catheter usage, urine leakage and a time consuming toilet routine are described. The subcategory bodily maturation describes that persons with spina bifida may experience an early bodily maturation, with puberty occurring at a younger age. The sensory perception subcategory contains descriptions of problems with balance and a deficient sense of touch and temperature:

> "I can easily get frostbite in the legs (. . .) if my legs are ice cold, I will not notice that."

Cognitive effects

The category "cognitive effects" is described in three subcategories. The subcategory mathematical ability contains descriptions of difficulties with mathematics in school. In some statements a connection is made between such cognitive dysfunctions and having spina bifida, while others do not make this association:

> "No, at least I have not thought about it [difficulties with mathematics] in that way, but I do not know. Anyway, from what I have understood, it [spina bifida] does not affect (. . .) how you think."

Difficulties with planning and initiating activities are described in the executive functions subcategory. The descriptions include for example difficulties with getting school home assignments done. A third subcategory describes cognitive effects connected to the memory domain:

> "Well, remembering and things like that. There are some stuff in the head also that you can have some difficulties with. But it is not only physical (laughter)."

Maintaining health

The category "maintaining health" is described in five subcategories. The subcategory toilet activities emphasizes the importance of proper bladder management to avoid risks associated with kidney damage and urinary tract infections:

> "Because otherwise I will get, I can more easily get bacteria. And then I get urinary infections and there can be kidney malfunction and stuff like that."

The second subcategory describes physical exercise as a way to increase the level of physical functioning and to avoid pain. The subcategory skin sore management includes descriptions of the health risks associated with skin sores, although no strategies for managing sores apart from avoiding them were observed:

> "If I get a sore for example on the foot or something, then it may take quite a long time before it heals and it is quite easy to get an infection in it."

Achievement of independence was mentioned as a precondition for being healthy as an adult. However, this subcategory does not include any statements that elaborate on the link between independence and health. The subcategory no specific activities contains utterances stating that no precautions are necessary for persons with spina bifida to maintain their health.

Perceptions of condition-related knowledge

Attitude to own knowledge

The category "attitude to own knowledge" is described in three subcategories. The subcategory awareness of disability contains descriptions of always having been aware of having spina bifida. The managing of spina bifida had constantly been present as a part of their daily life and thus reminding them of their condition. Participants also expressed that their awareness of certain aspects of spina bifida had developed in a stepwise manner.

In the subcategory assessment of knowledge state, participants described their knowledge state as incomplete, formulated for example as a lack of a "deeper knowledge". Descriptions of specific areas where they lacked knowledge are also present:

> "(. . .) So that you do not have spina bifida any longer and that you can make it go away (. . .) I would like to know that, if you can get well from it."

Some interviewees expressed an opinion on information acquisition implying that they had no need for additional information about anything related to spina bifida.

The subcategory also includes statements from participants being explicitly uninterested in spina bifida:

> "(. . .) I have looked a little into spina bifida, but not so very much. It is not a subject that is of interest to me, really."

Process of knowledge construction

The category "process of knowledge construction" is described in three subcategories. The subcategory initial information contains participants' descriptions of not remembering the initial information that they were given about spina bifida. Some interviewees had vague memories of being initially informed by their parents. The parents also played a role as providers of further information throughout the knowledge building process for some participants by talking about spina bifida at home with their children. This subcategory also contains expressions of rarely or never talking about spina bifida at home. Other providers of information that were mentioned include personnel within the health care system:

> "Well, I visit a doctor like once a year (. . .) We talk about it [spina bifida] and he kind of checks my muscles and things like that."

Some of the participants expressed that they did not remember the information that they had received in various contexts.

In the subcategory self-initiated information acquisition, the children and adolescents with spina bifida described that they had not attempted to inform themselves about their condition through reading books or searching for information on the internet. In contrast, some participants stated that they had accessed information about spina bifida via the internet or information material provided by the health care system. Taking an active part in an adolescent's own learning could also involve combining information from different sources to construct condition-related knowledge:

> "Well, at hospital visits they have told me a little (. . .) they have not gone into so much detail concerning the medical stuff, with nerves and those things. What I learnt about nerves, I actually learnt in school, in science class (. . .) we talked about the body, and I thought 'right that must be how it works' (laughter)."

Discussion

The aim of the study was to describe the condition-related knowledge among a group of children and adolescents with spina bifida in a Swedish context. The study uses a combination of an interview and a questionnaire to shed light on different aspects of condition-related knowledge. In contrast, prior studies have investigated such condition-related knowledge in other countries using either interviews (Dunn, McCartan, and Fuqua 1988; Erickson 1992; Feldman and Varni 1985) or questionnaires (Greenley et al. 2006; O'Mahar et al. 2010).

It seems that some participants in the present study associated spina bifida only with the physical effects of the condition and were not aware of the cognitive effects. An awareness of the cognitive manifestations might make persons with spina bifida better prepared and motivated to adopt strategies to handle the consequences of, for

example, memory problems. Adopting such strategies could be important for school work and help maintain proper catheterization procedures.

The participants were knowledgeable about the long term health risks associated with not adhering to the catheterization regime. These results are in contrast with previous studies. For instance, Erickson (1992) found that many young persons with spina bifida were relatively uninformed about precautionary measures such as bladder management. Greenley et al. (2006) found that children with spina bifida displayed inadequate knowledge about what happens if they forget to catheterize. Possibly, the advanced understanding among the study participants could be attributed to the use of a national follow-up programme in Sweden for people with spina bifida, focusing on neurogenic bowel and bladder dysfunction (Mattsson et al. 2009).

Some participants seemed to lack basic information about the function of the shunt and could not describe its connection to treatment of hydrocephalus. In addition, an inability to describe symptoms of shunt malfunctioning was found among many participants. Considering the potentially life-threatening consequences of shunt failure, knowledge about the symptoms is very important (Olsson et al. 2007). Greenley et al. (2006) reported similar findings concerning a lack of knowledge about signs of shunt malfunctioning. Another important aspect of maintaining health as persons with spina bifida reach adulthood is monitoring and managing sores (McDonnell and McCann 2000). Management of sores was mentioned in the interviews among the strategies and activities for maintaining health, however not by all participants.

The subject of etiology seemed to be challenging for some participants, and a lack of knowledge was evident. For example, it was not clear how the participants understood the concept of congenital conditions. It is possible that they acknowledged that the condition was congenital for them while at the same time not excluding that there could be other explanations to how a person contracts spina bifida. Contradictory views of spina bifida etiology could also be indicative of confusion between having spina bifida and being a wheelchair user. Indeed, a few participants seemed to equate spina bifida with movement impairments and assume that all persons with movement disabilities have spina bifida. A lack of basic understanding of the etiology of spina bifida among children and preadolescents with spina bifida was also observed by Greenley et al. (2006). Since the causative mechanism of spina bifida has not been fully established (Blom et al. 2006; Cabrera et al. 2004), questions regarding etiology could be expected to be difficult. Knowledge of the etiology of spina bifida might be important for understanding the permanence of the condition. In turn, this can have implications for developing realistic visions of their future lives and forming strategies to be used in their lives as adults.

Some of the participants that described their knowledge as incomplete or mentioned specific facts they did not know, also stated that they had no interest in acquiring information about spina bifida. Thus, the participants may consider their knowledge as adequate and may not feel a desire to learn more about their condition. In addition, they sometimes had not taken any initiatives themselves to find information about spina bifida. Such potential lack of motivation may be a complicating factor for any intervention attempting to develop the condition-related knowledge among persons with spina bifida. A previous intervention was not successful in increasing the condition-related knowledge of persons with spina bifida

(O'Mahar et al. 2010), although it is not clear whether this was related to a lack of motivation for learning among the participants.

Method discussion

The size of the participant group and the descriptive nature of the report limit the conclusions that can be drawn from the material, and therefore no generalizations to the whole population of persons with spina bifida in Sweden can be made. Given that spina bifida is a relatively rare condition (Josan, Morokoff, and Maixner 2008; Olsson et al. 2007), the number of potential participants for the study was expected to be small. Nevertheless, the persons invited for participation represent the entire population of eligible children and adolescents with spina bifida in a county in Sweden, and the level of non-participation was low. To facilitate transferability of the qualitative results, a detailed description of the constructed category system was provided together with example quotations. Attempts were made to increase the credibility of the interview data analysis by maintaining a dialogue between the researchers involved concerning the labelling and sorting of the data.

It was found that participants generally responded in a coherent way concerning the various knowledge domains across the two data collection instruments. However, the item *'Can people get spina bifida as adults?'* seemed to generate responses that were contradictory to interview statements. It is possible that the closed format of this question is not optimal since it does not give the participants a possibility to state if they think that there might be different reasons to why someone contracts spina bifida. For example, they may be aware that they contracted spina bifida congenitally while at the same time not excluding the possibility that adults can contract it if they fall in an accident. On some other areas of condition-related knowledge the participants revealed a more complex knowledge in response to the interview questions than probed by the questionnaire items. This highlights the benefit of using more than one type of data collection strategy when investigating condition-related knowledge.

Conclusions

The children and adolescents with spina bifida in this study had well-developed knowledge concerning proper bladder management, but were lacking knowledge about signs of shunt malfunctioning and etiology of spina bifida. Education about signs of shunt malfunctioning is crucial because children and adolescents will be required to take on an increasing responsibility for their own care and well-being as they grow older. A lack of awareness of health maintaining activities and the cognitive effects of the condition may be factors that impede the development of adequate strategies for dealing with their condition as they grow older. A potential confusion concerning the causes of spina bifida was observed, indicating that some participants have the incorrect view that spina bifida can be contracted by adults.

The interview and questionnaire gave a mostly coherent picture of this group's knowledge. This, in turn, suggests that the adapted test instrument (KOSB) employed in the present study can be an informative tool for health care personnel working with children and adolescents with spina bifida in Sweden. It might serve as a basis for the development of local clinical education programs in Sweden. However, if this instrument is to be used to assess the educational needs of persons with spina

bifida, it may be valuable to include additional health related factors such as sore management. Any educational intervention should take into account that persons with spina bifida do not necessarily have a motivation to learn more about their condition. Thus, work to increase motivation may be necessary before attempting to implement any educational strategies.

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