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Experiences among children and adolescents of living with spina bifida and their visions of the future

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Abstract

Purpose: Transitioning to independence may be problematic for persons with spina bifida (SB). Experiences of young persons with SB may provide insights into this group's needs for support. Therefore, the aim of this study was to investigate children's and adolescents' experiences of living with SB, their social and emotional adjustment, and their thoughts about becoming independent adults. *Method:* Semi-structured interviews were conducted with young persons with SB ($N=8$, age range 10–17 years). Social and emotional problems were assessed using Beck Youth Inventories. The interview transcripts were analysed using qualitative content analysis. *Results:* Three main themes were found: being a person with SB; everyday living as a person with SB; and preparing for life as an adult with SB. Indications of

emotional and social problems were most prominent among participants with milder physical disability. *Conclusions:* The findings indicate that young persons with SB may overestimate their independence. Other potentially problematic areas were lack of motivation, planning and preparedness for becoming independent. Research on transition to independence in this group should consider assistance at an early age in planning and executing strategies for independence. In addition, the potentially difficult situation for young persons with mild SB should be investigated further.

Keywords: spina bifida, independence, qualitative content analysis, children, adolescents, adjustment

Introduction

Spina bifida (SB) is a congenital birth defect that results in life-long physical and cognitive disabilities, as well as bladder and bowel dysfunction in most cases [1–3]. In addition, living with SB has also been associated with potential problems among children and adolescents with SB in achieving autonomy [4,5]. Problems persist into adulthood [6,7] and difficulties regarding employment and starting a family are not uncommon among persons with SB [8,9]. Facilitating the transition from adolescence to adulthood for persons with SB is becoming ever more important, given the increased survival rate into adulthood associated with advances in medical treatment [10]. Preparations for future life should start at an early age [11,12]. It is therefore crucial to develop intervention programs to support people with SB throughout the transition process [13], an endeavour that requires an understanding of young individuals' perspectives on living with SB [14].

Previous research has investigated several aspects of living with SB, including self-management [12,15], self-concept and family relationships [16], peer relationships [17],

experience of disability, self-understanding and identity [18,19], everyday living [20], faecal incontinence [21], social support [22], the process of transition to adult health care and adulthood [23,24] and condition-related knowledge [25]. The main focus of most studies is on how youth and adults with SB view their current situation rather than long-term plans and expectations.

Previous studies specifically focused on experiences and thoughts about the transition to adulthood and adult healthcare systems have focused on young adults with SB who are already in the midst of this process [23,24]. The findings describe participants' thoughts, in retrospect, regarding for example tensions between the comfort experienced in child health care and an emerging independence [23], and suggestions for what could have been done differently to facilitate the struggle for independence [24]. Other studies have targeted participants' experiences of self-management of the condition, an important part of transition. For example, in a study on adolescent women with SB, Sawin et al. [15] found that participants recognized deficits in self-management but few expressed plans for improvement. In addition, despite articulating goals of becoming independent, participants conveyed that they did not take advantage of opportunities to train self-management. Ito et al. [12] reported a variety of concerns and expectations for the future among adolescents and young adults with SB, including thoughts about employment and independent living, and worries about the development of the medical condition with age. Buran et al. [26] examined beliefs and expectations about living with SB, including future expectations, among adolescents. The participants were positive towards SB and optimistic about their future lives but were nevertheless found not to be very engaged in developing the skills necessary for transitioning to adulthood. The described literature appears to indicate a "gap" between the expressed goals and ambitions for the future among youth with SB and the fact that they may not actually initiate processes to attain these goals. However, research is needed to shed light

on how children and adolescents actually relate their current life situation to their aspirations for the future.

Attaining independent self-management of a chronic physical illness such as SB may be negatively affected by decreased psychological well-being. For example, Gadalla [27] observed an association between the presence of mood disorders and a less independent management of a chronic illness. In this regard, it should be noted that persons with SB are at a higher risk of developing symptoms of depression and anxiety as well as low self-esteem and a negative self-concept [28-32]. It is therefore of critical importance to consider the emotional and social functioning of children and adolescents with respect to their transition process (cf. [33]). There is to date not much research that relates experiences of living with SB to symptoms of social and emotional adjustment problems but there is related research focusing on aspects of identity and/or self-concept [16-19, 24, 34]. For example Bellin et al. described a mainly positive self-concept among young women with SB despite encountering stressors such as school related problems, episodes of teasing and bullying and strain resulting from the daily management of SB [16]. Ridosh et al. [24] have described experiences among young adults with SB of limiting their social interaction and being exposed to stigmatization such as bullying and social exclusion.

Studying not only how youth with SB experience living with the condition but also how they envision their adulthood may provide unique insights into how the process of becoming an independent adult with a high quality of life can be facilitated. As described above, there is a lack of research that investigates how children and adolescents with SB in early phases of their transition into adulthood view their adult life and what short-term strategies they employ in their striving for their long-term goals of becoming independent adults. Further, it is of interest to describe the social and emotional adjustment since this is a

factor that is relevant to consider when facilitating the transition to adulthood for this group. Therefore, the aim of the present study was to investigate children's and adolescents' experiences of living with SB, their social and emotional adjustment, and their thoughts about becoming independent adults.

Methods

The study design consisted of semi-structured interviews that were analysed using qualitative content analysis in combination with the Beck Youth Inventories (BYI) questionnaire [35]. The latter was used to complement the interviews with respect to the important domain of emotional and social adjustment.

Participants

The participants were Swedish children and adolescents aged 10–18 years with SB. The enrolment criteria required that the participants should not have intellectual disabilities more severe than the diagnosis of mild intellectual disability (i.e. ICD-10 code F70). Together with the lower age limit of 10 years, this criterion ensured that the participants were likely to understand the questions asked.

Recruitment took place at a clinic that monitors and manages the bowel and bladder functions of children and adolescents with SB [25]. Consent to participate was given by the persons with SB and their caregivers after being informed about the study. The recruitment efforts were stopped after eight interviews, at which time it was considered that the collected data reflected a range of experiences in a sample of children and adolescents that varied in age, sex and condition severity.

The background information about the participants was provided by a caregiver. Four females and four males aged from 10 to 17 years (median 14 years) participated (see table 1). Four walked without aid and the other four were wheelchair users and/or needed other assistive devices when walking. All except one used clean intermittent catheterization due to urinary bladder dysfunction and none of the participants were reported to have any intellectual disability. All participants were currently living with their parent/parents and were enrolled in school.

The regional research ethical committee of Linköping approved the study June 17, 2005 (M67-05).

Table 1. Key characteristics of participating children and adolescents with SB.

Participant code	Age	Sex	Ambulatory status	Deviating values on BYI ^a subscales
P1	10	female	Using ambulatory devices/wheelchair	-
P2	11	female	Walk without aid	Anxiety, depression, anger, disruptive behaviour, self-concept
P3	12	female	Walk without aid	Depression
P4	14	male	Using wheelchair	Anxiety
P5	14	female	Walk without aid	-
P6	15	male	Walk without aid	Anxiety, depression, anger, self-concept
P7	17	male	Using wheelchair	-
P8	17	male	Using wheelchair	-

^a BYI – Beck Youth Inventories

Data collection

The participants chose the location for the interview: at home (two participants) or at a local health care facility that they visited regularly (six participants). Only the participant and one researcher (author 1) were present during data collection. Efforts were made to ensure that the participant felt comfortable in the situation and was not distressed after completing the interview and questionnaire.

Interviews

Individual interviews were conducted during which the participants gave their views and experiences in the following three main areas: participants' 1) experiences of living with spina bifida, 2) ideas about future life, and 3) condition-related knowledge. The current study focus on results from the first two of these interview areas; findings regarding the participants' knowledge about SB has been reported previously [25]. A semi-structured interview format [36] was employed in which participants were posed a series of open-ended questions from an interview guide. Table 2 lists example questions for the two question areas. The interview guide was constructed based on the research aim, taking the available literature regarding the topic of transition to adulthood for persons with SB into account (in particular any identified relevant interview studies such as [15]) as well as knowledge regarding the language abilities of persons with SB [37]. During the interview, questions were rephrased if necessary to ensure that the interviewees understood the questions as intended, and follow-up questions were posed by the interviewer if required to elicit more detailed responses. The interviews lasted between 22 and 77 minutes and were audio recorded.

Table 2. Example interview questions for each of the investigated areas

Question areas	Example questions
Experiences of living with spina bifida	<p data-bbox="804 344 1286 367">What are your feelings about having spina bifida?</p> <p data-bbox="804 421 1390 472">In what ways are you similar to other youth your age that do not have spina bifida?</p> <p data-bbox="804 526 1366 577">What is important for you to consider when taking care of your body?</p> <p data-bbox="804 631 1203 654">Are there things that you need help with?</p>
Ideas about future life	<p data-bbox="804 703 1142 725">What are your plans for the future?</p> <p data-bbox="804 779 1318 801">Will you be able to manage on your own as an adult?</p> <p data-bbox="804 855 1366 907">What do you need to do now for you to become able to manage things independently when you become an adult?</p> <p data-bbox="804 960 1390 1012">Is there anything that you worry about regarding your life as an adult?</p>

Beck Youth Inventories

Given the importance of considering emotional and social adjustment in relation to the transition process for persons with SB and since children and adolescents may find it difficult to describe their psychosocial status in an open-ended interview format, the BYI questionnaire [35] was used as a complement to the interview. BYI has been translated and validated with a Swedish norm group consisting of 2358 children across the age range 9-18. The test exhibited excellent psychometric properties, with Cronbach alpha reliabilities between 0.89-0.94, and test-retest reliabilities ranging between 0.82-0.90. It consists of five self-reporting subscales in the domains of anxiety, depression, anger, disruptive behaviour and self-concept. Each subscale is composed of 20 items in the form of statements to which children and adolescents respond by rating how frequently each statement is true for them [35].

Data analysis

Interviews

Verbatim transcripts of the recorded interviews were subjected to content analysis [38] to discern ways in which the participants revealed their experiences. A qualitative approach was adopted that allowed categories to emerge from the data [39]. The analysis procedure (cf. [40]) commenced with gaining an initial sense of the transcribed interviews by reading through the material several times. Following this, the unit of analysis for the study was established by discerning and collating the parts of the transcripts that contained information about participants' perceptions of themselves and their current and future lives. The resulting text was then subdivided by defining meaning units consisting of complete or partial statements or a short sequence of statements. After condensing the meaning units, with appropriate consideration of the context, they were labelled with descriptive codes. Finally, meaningful structures and relationships in the data were identified and made explicit by sorting the codes and inductively building subcategories, categories and themes. Two of the researchers (author 1 and author 4) performed repeated cycles of analysis, discussion and re-analysis until there was agreement on the code labels, the sorting of codes into categories and formulation of the themes.

Beck Youth Inventories

The participants' responses to the BYI were scored as described in the manual [35]. BYI scores above the 75th percentile for the subscales of anxiety, depression, anger and disruptive behaviour and below the 25th percentile for the self-concept subscale are considered deviations and indicate potential social and emotional problems [35]. The BYI results were related to the interviews to reveal any differences in the experiences and views conveyed by persons with and without deviating BYI values. In addition, the distribution of

deviating BYI values in the group was compared with participants' mobility and sex characteristics to discern potential patterns.

Results

Interviews

Three main themes emerged from the qualitative content analysis of the interviews (table 3): “being a person with SB”, “everyday living as a person with SB”, and “preparing for life as an adult with SB”. In the following presentation, individual participants are referred to using the participant codes listed in table 1.

Table 3. Themes and associated categories and subcategories resulting from qualitative content analysis of the semi-structured interviews ($n=8$).

Categories	Subcategories
Theme 1: Being a person with SB	
Evaluating SB	Attitude to one's own disability Other people's responses
Evaluating oneself	Assessing one's competences Comparing oneself to others
Theme 2: Everyday living as a person with SB	
Consequences of having SB	Experiences of limitations Experiences of possibilities
Everyday strategies for handling one's disability	Self-controlled strategies Help-seeking strategies
Perceptions of one's own level of independence	Being an independent person Relying on help from other people

Theme 3: Preparing for life as an adult with SB

Making plans for the future	Thinking and talking about the future
	Hopes and expectations for the future
Considerations regarding future independence	Perceptions of level of independence in the future
	Strategies for becoming an independent person

Being a person with SB

This theme describes how the participants perceived themselves and how they related themselves to other people.

Evaluating SB

In this category, different attitudes towards their disabilities were expressed and experiences of how other people view and respond to their disabilities were described. The subcategory “attitude to one’s own disability” included participants’ expressions of sometimes conflicting emotions towards their disabilities. Attitudes were contingent on context in terms of locations and situations or what state of mind the participants were in. Negative emotional responses included sadness and irritability, whereas positive attitudes included attributing a wider perspective on life and an empowering stubbornness to living with SB. Positive attitudes could also arise from a sense of gratitude for having a relatively mild condition:

I think kind of positively, it [SB] is not a deadly disease and lots of people are worse off. Plus, most of the people with SB have problems walking, which I do not have. (P6, 15-year-old boy walking)

Neutral and accepting attitudes were also expressed, with participants viewing their disability as a part of ordinary life that they do not think about.

The subcategory “other people’s responses” included positive as well as negative reactions, and interviewees experienced support in the attitudes and behaviours of their peers and teachers. Negative responses ranged from intrusive questions about toilet regime to downright bullying behaviour by peers. In response to anticipated negative responses, a strategy of concealing the disability from others was verbalized by one interviewee:

The reason that I have not told my friends is that I see how they are now. Young people, and now I mean people my age, they are kind of, what can I say, immature or they say certain things. They think that if a person is different, then that person is a freak or something like that. That is why I have not dared [to tell them about having SB]. (P6, 15-year-old boy walking)

Evaluating oneself

In this category, the participants provided evaluations of their perceived abilities and related their own capacities to those of other people with and without SB. In the subcategory “assessing one’s competences”, the interviewees gave descriptions of both strengths and weaknesses, for example with respect to proficiency in school subjects and level of sports competence.

(laughter) I am quite good at exercising even though that I am not able to walk or run far, but I ride my bicycle. I bike a lot and kind of (pause) walk anyway (laughter). (P3, 12-year-old girl walking)

The subcategory “comparing oneself to others” included interviewees’ descriptions of similarities as well as dissimilarities to peers with and without SB. Although wheelchair users often gave general descriptions of sharing the same strengths and weaknesses as others with SB, the participants who could walk communicated more notions of dissimilarity, often by claiming that other persons with SB are more affected by their condition:

those [persons with SB] that I have met have had a lot more problems than me, with difficulties in walking at all (not audible), so I think I am one of those that are less affected. (P3, 12-year-old girl walking)

Some children and adolescents with SB perceived themselves as no different from people without SB. For example, one participant identified herself as being similar to peers without disabilities based on experiences when playing in a park area:

Well, I can join them. I can move pretty fast, although not as fast [as the peers]. (P1, 10-year-old girl using ambulatory devices/wheelchair)

The interviewees saw cognitive as well as physical differences between themselves and people without SB.

I probably do not get my homework done as fast as them [peers without SB] and I probably have more difficulties remembering things. And then there is the fact that you have to sneak out sometimes to go to the toilet. (P8, 17-year-old boy in a wheelchair)

Everyday living as a person with SB

This theme describes participants' everyday experiences of the effects of having SB, as well as their efforts to handle these consequences.

Consequences of having SB

In this category, the children and adolescents expressed the drawbacks and benefits associated with having SB. Their "experiences of limitations" included being limited in the range of accessible leisure activities and worrying about urine leakage and missing important lecture time in school because of toilet visits. Some interviewees said that their experiences of limitations were less severe after they achieved independent bladder management. Previously, they had been unable to stay over at a friend's house for example.

Although all participants experienced limitations arising from the toilet regime or impaired physical abilities, those who could walk expressed physical limitations with respect to specific activities, such as dancing and sports. In contrast, the limitations expressed by those who were unable to walk related more to basic functions, such as accessing stairs, walking, and keeping up with friends.

The subcategory “experiences of possibilities” included the opportunity to use wheelchair skills to impress peers by showing off. One participant pointed out that at least the wheelchair does not preclude spending a day in town with friends. Participants evaluated their possibilities optimistically by relating them to the fact that they could be worse off and that their problems could be more severe. For example, some interviewees expressed feelings of gratitude that they did not need a wheelchair, and some felt that the toilet regime was actually not so very cumbersome:

It [using the toilet] does not take more than five or six minutes, so I doubt that I miss very much [of lecture time]. (P8, 17-year-old boy using a wheelchair)

Everyday strategies for handling one’s disability

This category included the strategies that participants currently use to handle the effects of their disability, including maladaptive strategies associated with health risks. The subcategory “self-controlled strategies” included actively applied methods (e.g. using assistive devices) described by interviewees to overcome hindrances. Other strategies target cognitive functioning problems, such as using a calendar to aid memory and adapting school tasks.

The children and adolescents also described strategies to support more effective or independent management of their toilet regime. Other methods involved reducing the boredom associated with the sometimes time-consuming toilet regime by combining it with

doing something else as a distraction, such as playing a game. The maladaptive strategy of sometimes avoiding the toilet requirements was also expressed:

But sometimes maybe it... Sometimes I cannot bother to do it, and then I throw them [the catheters] away. (P2, 11-year-old girl walking)

Asking for assistance from others such as parents or teachers was expressed in the subcategory “help-seeking activities”. There were also expressions of reluctance to turn to other people for help because the person did not trust in other person’s ability to help.

Perceptions of one’s own level of independence

This category included descriptions of how participants experienced their current level of independence with regard to different aspects of their functioning. In the subcategory “being an independent person”, participants viewed themselves as essentially independent or requiring only limited support in school or from parents. Expressions of independence included cognitively managing the toilet schedule properly and initiating support strategies for planning in general. The adolescents revealed a sense of progressively increasing independence with time, primarily regarding the toilet:

...but when I was in pre-school the assistant even helped me with peeing and that kind of thing. But then when I started school I had to learn to do it myself, and then the assistant only gave support [with planning schoolwork for example]. (P5, 14-year-old girl walking)

The subcategory “relying on help from other people” included expressions across domains of life, for example being assisted by peers or parents when using public transport or reaching for objects at home. Assistance from teachers and/or assistants was needed during cognitive tasks such as remembering, planning and executing school tasks. Participants also expressed a need for help with the toilet schedule as well as with bowel management and in some cases with bladder management. Some participants conveyed contradictory views of

their level of independence. For example, a boy (P4) who initially claimed he had no need at all for assistance described a need for help from parents and assistants with fundamental aspects of functioning upon further probing later in the interview, including toilet management, school tasks, and accessing objects at home.

Preparing for life as an adult with SB

In this theme, descriptions of the participants' visions of how they will lead their lives as adults are presented.

Making plans for the future

This category includes the participants' hopes, expectations and plans for their future lives and their reflections on whether they had considered the time ahead of them. In the subcategory "thinking and talking about the future", none of the participants expressed that they had seriously considered their future, although some participants expressed unspecified worries regarding living with SB as an adult. Others were explicit in that they preferred not to think about the future and claimed they had no concerns for the future:

There are not a lot of things that can worry me. It is like, I improvise. If anything comes up that I cannot solve, we [participant and parents] will improvise. Then we will solve it when the problem arises. (P8, 17-year-old boy in a wheelchair)

Participants provided evaluations of the potential obstacles to specific ambitions (e.g. occupational) and areas of life caused by SB. None of the wheelchair users identified any specific hindrances, although one expressed an expectation of general problems related to accessibility. In contrast, most of the participants who could walk associated their disability with possible complications regarding starting a family and choosing an occupation.

The thing is that I am not good at mathematics and I have noticed that you should be [to manage the desired occupation] (laughter). I have some difficulties with mathematics, and then (...) I have difficulties remembering things, like I said, but I will probably have to... I have to practice so that I can remember (...) Writing things down, I have to pull myself together on that (laughter). It is this kind of thing that I might worry a bit about that it will not work out. (P5, 14-year-old girl walking)

In the subcategory “hopes and expectations for the future”, the children and adolescents described ideas about their adult life situation. The interviewees expected to move out of their parents’ home at various ages, typically around 20 years. Some wished to form a family and have children with a future partner.

Considerations regarding future independence

This category described the different ways that participants imagined themselves as independent adults and their strategies for overcoming limitations and achieving independence in the future. The subcategory “perceptions of level of independence in the future” included descriptions ranging from specific short-term goals, such as aiming for independent bowel management within 6 months, to general long-term expectations of becoming entirely independent as an adult. Participants who could walk expressed that they would be independent as adults, whereas, in contrast, most of the respondents in wheelchairs expressed more vague aspirations towards independence. Although the ambition was to become independent, they still allowed for unknown future problems that may require help from others, as exemplified by one boy’s reflection on whether he will be an independent adult:

Maybe not completely, but as good as...there is always something that you maybe will see as a problem later in life. I do not really know what now, actually. (P7, 17-year-old boy in a wheelchair)

The interviewees' expressed motivations for becoming independent included knowing that their parents wanted them to become independent. Not wanting to burden their parents was one source of motivation for managing the toilet routine by themselves. The children and adolescents offered vaguely articulated ideas about the extent of expected need of assistance in their daily life in the future (see quotation above) and who would provide the required help.

The subcategory "strategies for becoming an independent person" included participants' plans for how to overcome physical hindrances and to manage their planning and remembering. Participants also described that they did not know how to achieve the desired independence or that they found it difficult to get started. Interviewees harboured intentions to practice the toilet routine with the aim of being able to manage it independently in the future. However, some participants did not have clear ideas about what this practice might actually consist of:

Well, I have to do a bit of checking of the various ways you can do it [manage the toilet] and then practice the one [way of managing (the toilet)] that seems best, and it should be fine. (P7, 17-year-old boy in a wheelchair)

Beck Youth Inventories

Overall, half of the respondents (4 of 8) had deviating values compared with the norm group on at least one subscale each, indicating potential emotional or social problems. Two of the respondents who were walking without aid had deviating values on four (P6) and five (P2) of the BYI subscales, respectively (see table 1). Two other participants, one able to walk (P3) and one who used a wheelchair (P4), had deviating values on only one subscale each; the remaining respondents had no deviating values. Thus, a larger proportion of the participants who were able to walk without aid displayed deviating values compared with the

participants who needed aid in walking or were unable to walk (table 4). The distribution of deviating values was not different between males and females.

Table 4. Distribution of the results for the subscales of the Beck Youth Inventories for subgroups based on ambulatory status and sex. The numbers indicate the number of participants with deviating values on internalizing subscales (i.e. anxiety and depression), externalizing subscales (anger and disruptive behaviour), and the self-concept subscale. The number of participants with no deviating values is also shown.

Background variables	No deviating values	Anxiety and/or depression	Anger and/or disruptive behaviour	Self-concept
Ambulatory status				
Walk without aid (<i>n</i> =4)	1	3	2	2
Walk with aid or unable to walk (<i>n</i> =4)	3	1	0	0
Sex				
Female (<i>n</i> =4)	2	2	1	1
Male (<i>n</i> =4)	2	2	1	1

Utterances during the interviews stand out as possible reflections of emotional and social problems from the three individuals (P2, P3, P6) who were able to walk and had deviating values on the BYI subscales. Two of these participants (P2 and P3) expressed maladaptive, potentially destructive everyday strategies concerning the toilet during the interviews. Other verbal indications of psychosocial problems included negative emotional attitudes to SB in present time as well as negative reactions from other people and a perceived need to conceal the condition.

Discussion

The present study sheds light on children's and adolescents' experiences of living with SB, their social and emotional adjustment and their thoughts about becoming independent adults. As persons with SB transition from childhood to adulthood, problems in attaining skills of autonomy [4,5] and living independently as adults [6,8] may arise. Previous research has indicated a "gap" between the ambitions of becoming independent among young persons with SB and what steps they take to achieve this (e.g. [11,15,26]). The perspectives of children and adolescents in the early phases of their transition into adulthood presented in this study contribute to an understanding of some aspects of how this inconsistency between ambitions and actions may arise among youth with SB.

The findings indicate that the participants may not assess their level of independence accurately. Although some claimed to be basically independent, this initial impression had to be revised given their responses to questions about their independence in relation to specific activities. Similar findings have been reported in a study by Antle et al. [22] in which young people with SB perceived their level of tangible support to be minimal but more extensive support was revealed with further probing. The seemingly inaccurate perceptions of being independent may arise because the required support has been present throughout participants' entire lives and might therefore be invisible to them. This is supported by the study findings that descriptions of actively asking for help were rare. Thus, work aimed at increasing independence in this group of patients should consider the need to be specific and to assess not only whether persons are able to perform specific activities but also if they actually execute the tasks themselves.

The participants in the present study did not express strong personal motivations for becoming independent. When motivations were mentioned, it was based on a sense of

consideration for their caretakers. Thus, it seems that some children and adolescents may lack an internal drive to increase independence. In addition, although many participants expressed an intention to become independent, a detailed strategy for managing this step was lacking. These findings may be connected to executive dysfunction, which is common among people with SB [41]. A study of self-management among adolescent women with SB also found a lack of determination and formulated plans about how to become independent in [15].

The participants described a range of hopes and expectations for the future similar to what might be expected for young persons without SB, including getting a job and starting a family. Previous research studies have presented similar findings for youth with SB [34]. The findings from the present study further indicate that the participants were not very interested in thinking about their future in detail. In particular, some of the older participants expressed strikingly carefree attitudes towards their future considering the short time frame before taking on adult responsibilities of actively planning their lives and solving obstacles caused by their disability. Furthermore, many of the participants seemed to have vague ideas about the problems that may arise in their future life as adults with SB. The findings indicate that it might be necessary for another actor to initiate and support planning for the future. Ruck and Dahan-Oliel [11] have reported a similar lack of readiness for adulthood among adolescents and young adults with SB in terms of knowledge and skills needed to be independent, manage health care without help and access different community resources.

In the current study, it was particularly common that wheelchair users did not anticipate how their adult lives would be. Although they tended to describe current limitations in terms of basic functioning, they appeared not to extrapolate these problems into the future, and were less certain about achieving independence. In contrast, those who were able to walk seemed to have clearer ideas about becoming independent as adults; they

described current limitations in relation to specific activities and more often described potential obstacles in the future. Thus, they seemed to be able to relate their current situation to a possible future scenario for how their lives will evolve.

Psychological well-being may have an impact on successful transition to adulthood since it has been found to be related to self-management of chronic illness [27]. For example, Bellin et al. [33] found a negative correlation between symptoms of depression and self-management among young adults with SB. In the present study, more participants displayed deviating BYI scores on the subscales for depression and anxiety than on the subscales for anger and disruptive behaviour, and some participants displayed signs of a negative self-concept on the corresponding BYI subscale. This suggests that the participants tended to show more internalizing than externalizing symptoms, which is similar to findings reported in previous research [28,31,32,42].

The participants who were able to walk exhibited more indications of social and emotional problems compared with participants who were unable to walk. Similar findings have indicated that children and adolescents with functionally more severe SB have higher self-esteem [30] and higher health-related quality of life [43] than those with less severe disabilities. However, other studies have shown contrasting results [32,44]. The observed “condition-severity paradox” (cf. disability paradox [45,46]) may contradict intuitive expectation that a more severe disability would be correlated with more emotional and social problems. Müller-Godeffroy et al. [43] have suggested that observations whereby increased emotional problems are associated with a milder severity of SB may be understood through the concept of marginality.

Marginality in the context of disability is a state characterized by an ambiguous self-perception that includes aspects of an identity as a healthy person as well as a disabled person

[47]. Thus, children and adolescents with a less severe impairment may develop more problems with emotional and social adjustment because it is difficult for them to identify with healthy peers as well as with peers who are more severely impaired. In line with this reasoning, the present study revealed expressions that may indicate a state of marginality.

From the perspective of the marginality concept, the tendency of participants who can walk to focus on their differences from other persons with SB may be interpreted as indicating an ambiguous identity at the margin between disability and health. Those who used a wheelchair, in contrast, primarily identified similarities with other people with SB. Thus, those with SB who can walk may not identify with the physical disabilities that may result from SB, which may, in turn, result in more focus on trying to be similar to people who can walk. In addition, because their disabilities might not be directly observable by their peers, there may be more tension surrounding the differences in, for example, toilet routines. In this study, the participants with low levels of disability described negative reactions from other people, a factor that may limit the possibility of forming relationships with peers [24].

With an ambiguous identity at the margin between disability and health, people with mild SB may be more inclined to evaluate their competences in relation to people without disability. It seems likely that such comparisons could result in more feelings of disappointment and frustration [19]. In particular, it might be more difficult to maintain a positive outlook, which may be a protecting factor for people with SB [14]. This might be a more accessible strategy for those with a more severe disability. For example, aspects of functioning that people without disability would take for granted (e.g. meet friends in town) were seen as possibilities afforded by SB by some participants in this study who were unable to walk. The concept of marginality might be a fruitful framework for further research on a

larger sample that could disentangle the associations between condition-severity, psychological well-being, and development of independence among young persons with SB.

Method discussion

The main limitation of this study was the small number of participants, and it is likely that a larger sample could have further enriched the descriptions of experiences of living with SB. In addition, the wide age range in combination with the small sample size precluded an analysis of any developmental aspects of the experiences described by the participants. The findings nevertheless contribute unique insights into the views of children and adolescents with SB and what it can be like to live with the condition. In addition, the emerging patterns that were uncovered with respect to the development of independence and social and emotional adjustment has potential implications for practice and for future research. Transferability of the qualitative results was pursued by describing the resulting category system in detail and dialogue between the researchers was maintained throughout the categorization process to increase the credibility of the data analysis.

The data collection methods were chosen to be compatible with the age and cognitive characteristics of the participants. Thus, the decision to use a semi-structured interview format to help the interviewees keep on topic and formulate themselves within the different areas of interest in the study was based on the presumed language skill profile of the study group. The language abilities of children with SB has been described as referentially underspecified and characterized by a conversational style that contains non-relevant information [37]. The semi-structured interviews provided thorough descriptions from the participants on how they view themselves, their life situation and their thoughts about their lives as adults, indicating that this might be a suitable approach to data collection for this group. Using the BYI provided an indication of emotional and social adjustment problems in

addition to what was revealed during the interview. This form of method triangulation [48] gave opportunities to relate deviating values on the BYI to articulations of distress and potential stressors in the interviews for some participants. In addition, it was possible to relate a measure of psychosocial well-being to different aspects of the individuals' experiences of living with SB, with respect to for example the severity of the condition. The combination of questionnaire and interview used in this study provides unique opportunities to understand psychosocial aspects of living with SB, with potential clinical relevance for facilitating the transition process.

Conclusions

The results of this study indicate that children and adolescents with SB may overestimate their own level of independence. Although they have a desire to become independent, they nevertheless seem to lack a detailed plan of how to achieve this and a drive to work towards this goal. These aspects need to be considered when formulating health care programmes targeted to children and adolescents with SB. There is thus a need for the professionals who meet young people with SB to help them formulate and initiate strategies for achieving their short- and long-term goals. Furthermore, the results suggest a potential condition-severity paradox, which shows that it cannot be assumed that less severe levels of disability are always accompanied by a lower impact on the emotional and social adjustment of children and adolescents with SB. The findings imply that screening for problems with social and emotional adjustment should be part of routine follow-ups of individuals with SB, not least for those with minor ambulatory impairment.

Declaration of interest

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References

1. Dennis M, Barnes MA. The cognitive phenotype of spina bifida meningomyelocele. *Dev Disabil Res Rev* 2010;16:31-9.
2. Olsson I, Dahl M, Mattsson S, Wendelius M, Åström E, Westbom, L. Medical problems in adolescents with myelomeningocele (MMC): an inventory of the Swedish MMC population born during 1986-1989. *Acta Paediatr* 2007;96:446-9.
3. Oakeshott P, Hunt GM, Poulton A, Reid F. Open spina bifida: birth findings predict long-term outcome. *Arch Dis Child* 2012;97:474-6.
4. Davis, BE, Shurtleff DB, Walker WO, Seidel KD, Duguay S. Acquisition of autonomy skills in adolescents with myelomeningocele. *Dev Med Child Neurol* 2006;48:253-8.
5. Friedman, D, Holmbeck GN, DeLucia C, Jandasek B, Zebracki K. Trajectories of autonomy development across the adolescent transition in children with spina bifida. *Rehabil Psychol* 2009;54:16-27.
6. Jenkinson, MD, Campbell S, Hayhurst C, Clark S, Kandasamy J, Lee MK, Flynn A, Murphy P, Mallucci CL. Cognitive and functional outcome in spina bifida–Chiari II malformation. *Childs Nerv Syst* 2011;27:967-74.
7. Hunt GM, Oakeshott P. Lifestyle in adults aged 35 years who were born with open spina bifida: prospective cohort study. *Cerebrospinal Fluid Res* 2004;1:4.

8. Vu Minh Arnell M, Svedberg KS, Lindehall B, Jodal U, Abrahamsson K. Adults with myelomeningocele: an interview study about life situation and bladder and bowel management. *J Pediatr Urol* 2013;9:267-71.
9. Barf HA, Post MWM, Verhoef M, Jennekens-Schinkel A, Gooskens, RHJM, Prevo AJH. Life satisfaction of young adults with spina bifida. *Dev Med Child Neurol* 2007;49:458-63.
10. Davis BE, Daley CM, Shurtleff DB, Duguay S, Seidel K, Loeser JD, Ellenbogen RG. Long-term survival of individuals with myelomeningocele. *Pediatr Neurosurg* 2005;41:186-91.
11. Ruck J, Dahan-Oliel N. Adolescence and young adulthood in spina bifida: self-report on care received and readiness for the future. *Top Spinal Cord Inj Rehabil* 2010;16:26-37.
12. Ito JA, Stevenson E, Nehring W, Alpeter A, Grant J. A qualitative examination of adolescents and adults with myelomeningocele: their perspective. *Eur J Pediatr Surg* 1997;7(S1):53-54.
13. Binks JA, Barden WS, Burke TA, Young NL. What do we really know about the transition to adult-centered health care? A focus on cerebral palsy and spina bifida. *Arch Phys Med Rehab* 2007;88:1064-1073.
14. Lindsay S. A qualitative synthesis of adolescents' experiences of living with spina bifida. *Qual Health Res* 2014;24:1298-309.
15. Sawin KJ, Bellin MH, Roux G, Buran CF, Brei TJ. The experience of self-management in adolescent women with spina bifida. *Rehabil Nurs* 2009;34:26-38.

16. Bellin MH, Sawin KJ, Roux G, Buran CF, Brei TJ. The experience of adolescent women living with spina bifida part I: self-concept and family relationships. *Rehabil Nurs* 2007;32:57-67.
17. Roux G, Sawin KJ, Bellin MH, Buran CF, Brei TJ. The experience of adolescent women living with spina bifida part II: peer relationships. *Rehabil Nurs* 2007;32:112-19.
18. Kinavey C. Explanatory models of self-understanding in adolescents born with spina bifida. *Qual Health Res* 2006;16:1091-107.
19. Kinavey C. Adolescents born with spina bifida: experiential worlds and biopsychosocial developmental challenges. *Issues Compr Pediatr Nurs* 2007;30:147-64.
20. Fägerskiöld AM, Mattsson GG. Disabled children and adolescents may be outsiders in the community. *Int Nurs Rev* 2010;57:470-7.
21. Johnsen V, Skattebu E, Aamot-Andersen A, Thyberg M. Problematic aspects of faecal incontinence according to the experience of adults with spina bifida. *J Rehabil Med* 2009;41:506-11.
22. Antle BJ, Montgomery G, Stapleford C. The many layers of social support: capturing the voices of young people with spina bifida and their parents. *Health Soc Work* 2009;34:97-106.
23. Garibaldi KL, Gibson RW, Reiss J, Villarreal G B, Haidet P. Transition from pediatric to adult healthcare: the experiences of young adults with spina bifida. *J Gen Intern Med* 2004;19(Suppl S1):229.
24. Ridosh M, Braun P, Roux G, Bellin M, Sawin, K. Transition in young adults with spina bifida: a qualitative study. *Child Care Health Dev* 2011;37:866-74.

25. Strömfors L, Falk L, Wilhelmsson S, Höst GE. Condition-related knowledge among children and adolescents with spina bifida in a Swedish county. *Scand J Disabil Res* 2014;16:127-40.
26. Buran CF, Sawin KJ, Brei TJ, Fastenau PS. Adolescents with myelomeningocele: activities, beliefs, expectations, and perceptions. *Dev Med Child Neurol* 2004;46:244-252.
27. Gadalla T. Association of comorbid mood disorders and chronic illness with disability and quality of life in Ontario, Canada. *Chronic Dis Can* 2008;28:148-54.
28. Ammerman RT, Kane VR, Slomka GT, Reigel DH, Franzen MD, Gadow KD. Psychiatric symptomatology and family functioning in children and adolescents with spina bifida. *J Clin Psychol Med Settings* 1998;5:449-65.
29. Appleton PL, Minchom PE, Ellis NC, Elliott CE, Böll V, Jones P. The self-concept of young people with spina bifida: a population-based study. *Dev Med Child Neurol* 1994;36:198-215.
30. Minchom PE, Ellis NC, Appleton PL, Lawson V, Böll V, Jones P, Elliott CE. Impact of functional severity on self concept in young people with spina bifida. *Arch Dis Child* 1995;73:48-52.
31. Shields N, Taylor NF, Dodd KJ. Self-concept in children with spina bifida compared with typically developing children. *Dev Med Child Neurol* 2008;50:733-43.
32. Bellin MH, Zabel TA, Dicianno BE, Levey E, Garver K, Linroth R, Braun, P. Correlates of depressive and anxiety symptoms in young adults with spina bifida. *J Pediatr Psychol* 2010;35:778-89.

33. Bellin MH, Dosa N, Zabel TA, Aparicio E, Dicianno BE, Osteen P. Self-management, satisfaction with family functioning, and the course of psychological symptoms in emerging adults with spina bifida. *J Pediatr Psychol* 2013;38:50-62.
34. Wollenhaupt J, Rodgers B, Sawin KJ. Family management of a chronic health condition: perspectives of adolescents. *J Fam Nurs* 2012;18:65-90.
35. Beck JS, Beck AT, Jolly JB. Beck youth inventories manual Swedish version. Translated by Tideman E. Stockholm: Psykologiförlaget; 2004.
36. Kvale S. *Interviewing: learning the craft of qualitative research interviewing*. Los Angeles: Sage Publications; 2009.
37. Dennis M, Landry SH, Barnes M, Fletcher JM. A model of neurocognitive function in spina bifida over the life span. *J Int Neuropsychol Soc* 2006;12:285-96.
38. Krippendorff K. *Content analysis: an introduction to its methodology*. Beverly Hills: Sage; 1980.
39. Hsieh HF, Shannon SE. (2005). Three approaches to qualitative content analysis. *Qual Health Res* 2005;15,1277-1288.
40. Graneheim UH, Lundman B. Qualitative content analysis in nursing research: concepts, procedures and measures to achieve trustworthiness. *Nurs Educ Today* 2004;24:105-12.
41. Tuminello ER, Holmbeck GN, Olson R. Executive functions in adolescents with spina bifida: relations with autonomy development and parental intrusiveness. *Child Neuropsychol* 2012;18:105-24.
42. Appleton PL, Ellis NC, Minchom PE, Lawson V, Böll V, Jones P. Depressive symptoms and self-concept in young people with spina bifida. *J Pediatr Psychol* 1997;22:707-22.

43. Müller-Godeffroy E, Michael T, Poster M, Seidel U, Schwarke D, Thyen U. Self-reported health-related quality of life in children and adolescents with myelomeningocele. *Dev Med Child Neurol* 2008;50:456-61.
44. Hommeyer J, Holmbeck G, Wills K, Coers S. Condition severity and psychosocial functioning in pre-adolescents with spina bifida: disentangling proximal functional status and distal adjustment outcomes. *J Pediatr Psychol* 1999;24:499-509.
45. Albrecht GL, Devlieger PJ. The disability paradox: high quality of life against all odds. *Soc Sci Med* 1999;48:977-88.
46. Drum CE, Horner-Johnson W, Krahn GL. Self-rated health and healthy days: examining the “disability paradox”. *Disabil Health J* 2008;1:71-8.
47. Friedman SB. The concept of "marginality" applied to psychosomatic medicine. *Psychosom Med* 1988;50:447-53.
48. Merriam S. B. *Qualitative research: a guide to design and implementation*. San Francisco: Jossey-Bass; 2009.