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Research Paper

Outcome and life satisfaction of adults with myelomeningocele

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Abstract

Background: Myelomeningocele (MMC) commonly causes impairments in body structure and functions as well as cognitive disabilities that can have an adverse effect on adult life. Improved medical care has resulted in increased numbers of individuals with MMC surviving to adulthood, however little is known about the impact of MMC on the lives of adults age 25 years or older.

Objective: To gain a better understanding of outcomes in education, employment, relationships, reproduction and life satisfaction of adults with MMC.

Methods: A primarily quantitative multiple-choice questionnaire designed to capture outcomes in education, employment, relationships and reproduction, along with a previously validated life satisfaction checklist (LiSat-11), was completed by adults with MMC. Relationships between demographic variables, outcomes and life satisfaction were determined using cross tabulation analysis, logistic regression and linear regression.

Results: Ninety adults with MMC, age 25−85 years (median age 32), reported a diverse range of outcomes in education, employment, relationships and reproduction. The most consistent variable associated with difficulty attaining adult milestones was hydrocephalus, the presence of which reduced the likelihood of living independently ($p \leqslant 0.001$), having a partner ($p = 0.003$) and reproducing ($p \leqslant 0.001$), but did not contribute to reduced life satisfaction.

Conclusions: Adults with MMC, especially those without hydrocephalus, can obtain gainful employment, live independently, form partner relationships and have children, and these achievements contribute to life satisfaction. While MMC does not affect overall reported life satisfaction for adults, attention should be paid to specific domains with less reported satisfaction.

Keywords: Myelomeningocele; Spina bifida; Adults; Life satisfaction

Myelomeningocele (MMC), the most common type of spina bifida, is a neural tube defect (NTD) resulting from failure of the caudal end of the neural tube to close by roughly day 28 post ovulation.[1,2] With the introduction of immediate spinal closure and shunting for hydrocephalus in newborns with MMC in the 1960s, the survival rate increased dramatically from approximately 10% to an estimated 46−75% of individuals with MMC surviving into adulthood today.[3−7] Nearly 25,000 children and adolescents with spina bifida were estimated to be living in the United States in 2002, the majority of whom will reach adulthood in the coming years.[8]

Despite improved medical treatment, individuals with MMC often have lasting cognitive disabilities, impairments in body structure such as scoliosis, and impairments in body functions including muscle weakness, paralysis and bowel and bladder incontinence, all of which can have an adverse effect on adult life.[5,9] Predictors of long-term outcome, such as hydrocephalus, continence, lesion level, IQ, gender and mobility have been investigated in European and North American populations, though previous

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Abstracts of the data have been presented at the NSGC 30th Annual Education Conference and the 7th International Conference on Neural Tube Defects. A brief synopsis of results was posted on the Duke Center for Human Genetics website as part of a newsletter for study participants.

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research has been limited and results are conflicting.[5,6,10–16] The presence of hydrocephalus, which occurs in approximately 80% of individuals with MMC, significantly increases the likelihood of cognitive impairment and is the most commonly reported variable associated with poor outcome.[10–13,17–19]

The vast majority of MMC outcome research to date has been conducted either with populations of young adults, a distinct developmental group between the ages of 18 and 25 years, or in broad populations consisting of adolescents, young adults and adults. While these data are informative, these studies are not readily transferrable to adults with MMC. Adolescents and young adults may not have had adequate time to achieve life goals, have less medical complications and better preserved mobility than adults.[3,6,12,14,19–22] Impairments in body structure and functions such as scoliosis, renal damage and pressure sores often worsen with age and the likelihood of independent mobility decreases, which may reduce the ability of adults to actively participate in society.[23–25] After extensive literature review, only three outcome studies were identified in which the study population consisted exclusively of adults with MMC, defined as individuals age 25 years or older.[4,6,25] The purpose of this study is to gain a better understanding of outcomes in education, employment, relationships, reproduction and life satisfaction of adults with MMC.

As individuals with MMC reach adulthood, they face challenges in attaining milestones commonly used to indicate positive outcomes, such as obtaining postsecondary education, employment, living independently and forming partner relationships.[14,26] Previous research indicates that a reduced number of young adults with MMC pursue postsecondary or graduate education.[7,11,13,20,27] Individuals with MMC also have significantly higher unemployment rates than the general population and those who are employed tend to hold low-wage jobs.[4,6,7,13,14,17,20,28,29]

Individuals with MMC report a typical desire to live independently, have romantic relationships, get married and even have children.[16,19,21,28] However, research has shown that the majority of individuals with MMC require some form of living assistance, have difficulty forming romantic relationships and have low rates of marriage.[4,6,7,13,14,16,19,28,30,31] Though the majority of women with MMC do not have children, there is no evidence suggesting decreased fertility in this population.[4,6,20,31,32] Women with MMC have normal menstruation, are able to become pregnant and have healthy babies, however, unique concerns for these women have been reported including recurrent urinary tract infections, worsening kidney function, premature labor and postoperative complications.[33,34] In contrast to women, data show that men with MMC have reduced fertility but a proportion, typically those with lower level lesions, can successfully reproduce.[30,35] Additionally, both men and women with MMC are at increased risk to have a baby with an NTD.[34,36] Preconception folic acid supplementation may reduce this risk by up to 70%.[37]

Despite physical burdens and social difficulties, research suggests that young adults with MMC are as satisfied with their lives as typically developing peers.[12,20,38] However, certain aspects such as finances, self-care ability and partner relationships are often reported to be less satisfying.[12,15,19,23] Unfortunately, life satisfaction research on adults with MMC is minimal, which leads to unclear expectations for the future.[14,39]

Methods

Participants

Participants were recruited from the Hereditary Basis of Neural Tube Defects study conducted at the Center for Human Genetics at Duke University Medical Center.[36] Ascertainment of participants occurred across the United States from 1994 until 2007 from a variety of sources including spina bifida clinics, support groups and self-referral in response to advertisements. Adults age 25 years or older with a reported diagnosis of MMC were contacted for participation in this study.

From a pool of over 5000 participants, 325 were eligible to participate in this study. Consent was obtained from 124 individuals (38%) and 99 (30%) completed the questionnaire. Nine participants were excluded for reporting a diagnosis other than MMC, leaving 90 for analysis. There were no significant differences between responders and non-responders in regards to gender, race, age, lesion level or hydrocephalus status. IRB approval was obtained from Duke University Medical Center, as well as the University of North Carolina at Greensboro.

Measures

A primarily quantitative, 77-item multiple-choice questionnaire, which consisted of four outcome sections (education, employment, relationships and reproduction) was developed by the authors using published literature[10–13,16,17,20,21,31] and previously developed questionnaires obtained from other research groups.[6,29,40] Qualitative items were used to capture postsecondary degrees, job titles, partner disabilities and pregnancy complications. Additionally, participants were asked to describe why they were or were not concerned about having a child with an NTD. The questionnaire was pilot-tested on two adults with MMC and five health care professionals who either had experience caring for patients with MMC or in questionnaire development.

The LiSat-11, a self-administered life satisfaction checklist, was used to correlate outcomes in education, employment, relationships and reproduction to life satisfaction. Participants rated satisfaction with 10 life domains (life as a whole, employment, finances, leisure, contact with

friends and acquaintances, self-care, family life, partner relationships, physical health, and mental health) along a six-point sale from 1 (very dissatisfying) to 6 (very satisfying). The sex life domain was not included. The LiSat-11 was previously validated in a population of randomly selected Swedish adults, from which reference values exist.[41]

Procedure

Participants completed the questionnaire either online or via mailed paper copy. Medical records were used to confirm the diagnosis of MMC, lesion level and presence or absence of hydrocephalus. If medical records and patient report contradicted, data from medical records were used. Medical records were not available on all participants ($n = 24$, 27%) and, in those cases, patient report was not verified. Qualitative items were coded into categories by two independent researchers. Coding disagreements were resolved through discussion with a third researcher until a consensus agreement was reached. Job titles were coded based on U.S. Bureau of Labor Statistics categorizations.[42]

Statistical methods

Data analysis was completed in SAS version 9.2 (SAS Systems, Cary, NC). Frequencies of each variable were calculated and summarized (PROC FREQ). Cross tabulation analysis was then used to determine the relationships between outcome and different variables (gender, lesion level, age (born before or after 1960), hydrocephalus, continence and mobility) in each section. Chi-square tests were used to determine statistical significance between these differences. Analysis of LiSat-11 was done using previously reported methods.[41] Domain scores were summed and an average score was calculated for each. Analysis of Variance (PROC GLM) was used to determine significant differences in life satisfaction domains between demographic data and other variables.

To determine the effect, if any, of co-variance in the models, several variables were considered (age, gender, hydrocephalus, mobility, continence and lesion level). Bivariate regression was used to assess the association among these potential predictors and to limit collinearity. Consequently, age, mobility and lesion level were not included as they were all significantly associated with hydrocephalus. Multivariate analysis was subsequently performed with gender, hydrocephalus and continence included as co-variates. Logistic regression (PROC LOGISTIC) was employed for analysis of categorical outcomes while linear regression (PROC GLM) was used for continuous outcomes. A p value of <0.05 was used to determine statistical significance for all data analyzed. Unless stated, reported p values were derived from univariate analyses which remained significant after controlling for co-variates.

Results

Most participants completed the questionnaire themselves (88%) with the remainder completed by a parent (12%). All questions were voluntary; therefore the sum does not equal 90 for all items.

Demographics

Demographic characteristics of the study group are summarized in Table 1. Participants were 25−85 years old with a median age of 32 years. Participants were primarily white and evenly split between male and female. The majority of participants had lumbo/sacral lesions and required a shunt for the treatment of hydrocephalus. Twelve participants (13%) were born before 1960, none of whom had hydrocephalus. When controlling for co-variates such as hydrocephalus, the only difference between participants born before and after 1960, was a greater likelihood of the older group having children. Otherwise, there were no differences in outcomes based solely on age. Nearly half of all participants required a wheelchair for mobility. Most participants did not have complete bowel and bladder continence (defined as less than one bowel or bladder accident per month).

Education

Table 2 summarizes the highest level of education obtained for the study group. The vast majority of participants (94%) completed high school or a high school equivalent (GED). The majority of participants (62%) completed their high school education in a typical classroom setting, 13% required an aide or extra one-on-one assistance, 10%

Table 1
Study participant demographics ($N = 90$)

	n	%
Sex		
Female	50	56
Male	40	44
Race		
White	84	93
Black	1	1
Other	5	6
Mobility ($N = 88$)		
Wheelchair	42	48
Independent	24	27
Assistive devices	22	25
Highest lesion level		
Lumbo/sacral	77	86
Thoracic	13	14
Hydrocephalus		
Yes	63	70
No	26	29
Unsure	1	1
Continence		
No	70	78
Yes	17	19
Unsure	3	3

Table 2
Highest completed education ($N = 90$)

	n	%
No formal schooling	3	3
Some high school	2	2
High school/GED	26	29
Some college	9	10
Technical or associates degree	16	18
College degree	30	33
Advanced/graduate degree	4	4

required special education for specific subjects only, and 14% were primarily in a special education classroom. Participants with hydrocephalus were less likely to be in a typical classroom setting in high school ($p = 0.007$). There was no association between gender, continence, or mobility and high school educational setting.

Over half of participants (56%) completed a technical, associates or college degree. The majority (44%) obtained a degree in the social sciences, with the remainder obtaining degrees in business (20%), humanities (17%), natural or medical sciences (12%), engineering or computer science (5%) and general education (2%). Participants in a typical classroom setting in high school were more likely to obtain postsecondary education ($p = 0.0001$). There were no significant associations between hydrocephalus, lesion level, mobility, gender, age or continence and obtaining postsecondary education.

Employment

Participants primarily received income from disability benefits (59%) and/or their own employment (48%). The majority of participants (85%) held a job at some point during their life with just under half (48%) currently employed. Participants reported a wide variety of job titles including front desk clerk, medical transcriptionist, cashier, social worker, certified public accountant (CPA) and elementary school teacher. Job categories were as follows: administrative support (28%), professional and technical (24%), executive, administrative, and managerial (11%), handlers, equipment cleaners, helpers and laborers (11%), service (10%), sales (10%), precision production, craft, and repair (4%), and transportation and material moving (1%).

Table 3 summarizes key characteristics of participants' current or most recent jobs. Approximately half of working participants (51%) worked part-time (less than 40 h per week). The most common reported reasons for part-time employment included health issues (42%) and job availability (35%). Most participants earned a salary of less than $25,000 per year, which is below the national average of $29,730.[43] There was no association between gender and income. There was also no association between employment and gender, age, mobility, lesion level, continence, or hydrocephalus.

Reported reasons for current unemployment included being laid off (40%), resigning (30%), retiring (15%) and being fired (9%). A total of 17% of participants never held a job. Of those participants, only one was currently searching for employment. Of participants who never held a job, 82% attributed this to physical or medical limitations and 45% cited learning disabilities.

Relationships

Table 4 summarizes the living situation of the study group. Nearly half (43%) of participants still lived with their parents and 21% lived alone with or without assistance. Individuals who were independently mobile ($p = 0.003$) and those without hydrocephalus ($p = 0.0001$) were more likely to live independently (alone or with a partner, spouse or roommate). There was no association between participants' living situations and continence, lesion level, age or gender.

Table 4 also summarizes participants' romantic relationship histories. Over half (52%) of participants never had a serious partner or spouse, while 28% had been married. While the rate of marriage was low, for those who did marry, the age of marriage was consistent with the general population.[44] Seven participants (17%) reported having a partner with a disability including NTDs, blindness and cerebral palsy. Individuals without hydrocephalus ($p = 0.003$) and with independent mobility ($p = 0.001$) were more likely to be in a relationship. There was no association between relationship status and age, gender, lesion level or continence.

Of participants who never had a partner, 20% reported a desire for a serious partner or spouse someday while 64% were unsure.

Table 3
Job characteristics (current or most recent)

	n	%
Number of years at job ($N = 74$)		
<1 year	19	26
1–5 years	27	36
6–10 years	15	20
>10 years	13	18
Hours worked per week ($N = 73$)		
1–9 h	10	14
10–19 h	10	14
20–29 h	14	19
30–39 h	3	4
40 h	23	31
>40 h	13	18
Salary ($N = 71$)		
<$25,000 per year	51	72
$25–50,000 per year	12	17
$51–75,000 per year	4	6
$76–100,000 per year	4	6
>$100,000 per year	0	0

Table 4
Participants' living situations and relationship histories

	n	%
Living situations ($N = 88$)		
Alone without assistance	12	14
Alone with assistance	6	7
Partner, spouse, roommate or friend	27	31
Parents	38	43
Group home	5	6
Current relationship status ($N = 87$)		
Never had a partner	45	52
Previous partner, never married	8	9
Current partner, not married	10	11
Married	16	18
Separated/divorced	6	7
Widowed	2	2
Age at marriage ($N = 25$)		
<20 years old	1	4
20–25 years old	16	64
26–30 years old	6	24
31–34 years old	1	4
35–40 years old	1	4
Where met partner/spouse ($N = 43$)		
Church, community group, club, or social event	12	28
Through family member or friend	10	23
School	4	9
Work	7	16
Internet	8	19
Spina bifida support group	2	5
Reason for lack of partner ($N = 44$)[a]		
Not interested	6	14
Still looking	15	34
Physical disabilities	25	57
Learning disabilities	5	11
Not sure	12	27

[a] Could choose more than one answer.

Table 5
Participants' reproductive histories

	n	%
Biologic children ($N = 89$)		
Yes (men)	8	9
Yes (women)	8	9
No	73	82
Age at first biologic child ($N = 16$)		
<20 years old	0	0
20–25 years old	5	31
26–30 years old	8	50
31–34 years old	1	6
35–40 years old	2	13
First pregnancy full term ($N = 8$)[a]		
Yes	4	50
No	4	50
First pregnancy delivery ($N = 8$)[a]		
Vaginal	6	75
Planned C-section	2	25
Had baby with NTD ($N = 16$)		
Yes	3	19
No	13	81

[a] Females only.

Reproduction

Table 5 summarizes participants' reproductive histories. Sixteen participants (18%) had biologic children. All but one participant with biologic children, including all eight men, had lumbo/sacral lesions. Of the resulting 34 children, three (9%) had an NTD, born to two women and one man. This observed recurrence of NTDs in offspring is above the empiric recurrence risk due to recruiting participants from an NTD genetics study. The majority (80%) of participants (females or female partners of male participants) with biologic children born after the benefits of folic acid were known did take some amount of folic acid prior to pregnancy, either as part of a prenatal vitamin or alone.[37] Yet only two participants (13%) reported taking the therapeutic 4 mg dose. Participants with biologic children were more likely to be born before 1960 ($p \leqslant 0.001$) and less likely to have hydrocephalus ($p \leqslant 0.001$). No associations were found between biologic children and gender, mobility or continence.

The majority of female participants (71%) were concerned about the effects of pregnancy on their own health. Nearly half (43%) of female participants with biologic children experienced health or medical complications as a result of pregnancy or delivery. Medical complications included urinary tract and kidney infections and increased difficulty with mobility and continence. Of participants with biologic children, two men (25%) and two women (25%) reported difficulty conceiving. Two additional women (20%) and one man (11%) attempted to conceive without success. One participant adopted her spouse's children from a previous relationship.

Of participants with no biologic children, reported reasons for not having children were not having a partner/spouse (63%), choice (38%), not able to raise a child due to physical or learning disabilities (15%) and infertility (7%). Nearly half (41%) reported a desire to have biologic children in the future.

Over half (57%) of participants were concerned about having a child with an NTD. Concerned participants fell into two categories; those that didn't feel physically capable of caring for a child with a disability and those that didn't want their child to suffer as they had suffered. Participants who were not concerned (43%) also fell into two categories. Unconcerned participants either felt that the risk to have a child with an NTD was low or that their own experiences living with MMC prepared them to parent a child with an NTD.

Life satisfaction

Table 6 summarizes LiSat-11 mean domain scores. Overall, satisfaction with self-care and family life were the highest, while satisfaction with partner relationships, finances and employment were the lowest. When compared to reference values from an adult population, participant satisfaction with life as a whole was equivalent.[41] However, the domains of employment, contact with friends and acquaintances, self-care, partnership relationships, physical

Table 6
LiSat-11 mean domain scores[a] and factors associated with increased domain life satisfaction as determined by logistic regression

Domain	Mean	*N*	SD	Factors increasing satisfaction	*p* value
Family life	5.0	88	1.2	Hydrocephalus	0.02
Self-care	4.8	86	1.3	Postsecondary education	<0.001
				Living alone	0.01
				Continence	0.03
				Current or previous relationship	0.05
Life as a whole	4.7	87	1.3	Living with someone	0.005
Leisure	4.7	87	1.2	Hydrocephalus	0.05
Mental health	4.4	88	1.4	None	–
Friends	4.2	87	1.5	None	–
Physical health	4.1	88	1.3	Continence	0.03
Employment	3.9	84	1.7	Current employment	0.008
Finances	3.8	87	1.5	Current employment	0.03
Partner	3.5	74	2.0	Current or previous relationship	<0.001
				Independent mobility	0.01

[a] Scale of 1−6 with 1 being very dissatisfying and 6 being very satisfying.

health and mental health were all rated significantly lower than the reference group.

Associations between participant characteristics and increased domain scores are reported in Table 6. No differences in life satisfaction were seen based on gender, lesion level or age.

Discussion

Participants were exclusively adults age 25 years or older with MMC with a median age of 32 years; which is a more homogeneous adult population than has often been previously described.[3,12,14,15,19–23,38] The presence of hydrocephalus and lack of independent mobility contributed to difficulty attaining adult milestones while lesion level, gender and continence were not determining factors, which is consistent with some previous reports of young adults.[10,11,13,15,19] Recent research demonstrated the effectiveness of in-utero surgical repair in reducing rates of hydrocephalus and improving motor outcomes.[45] As these two variables seem to have the greatest impact on adult outcome, additional research into in-utero repair is warranted as the potential exists to significantly improve outcomes for future generations of adults with MMC.

In contrast to previous reports, when compared to the general population of adults 25 years of age and older in the United States, adults with MMC in the study group were more likely to complete high school, obtain technical or associates degrees and graduate college.[46] This could partly be explained by the young age of individuals included in previous research, who may not have had adequate time to complete their education. Individuals who received special education in high school were less likely to pursue education beyond high school, indicating that this group may be in particular need of assistance with obtaining postsecondary education. Participants who pursued postsecondary education often obtained degrees in the social sciences or humanities. Learning disabilities, specifically in problem solving and mathematics, in individuals with MMC has been reported; which may account for the lower number of degrees in these areas.[47] To optimize educational success, it may be advantageous for individuals with MMC to recognize common cognitive strengths and focus postsecondary education efforts in these areas.

Participants in the study group reported diverse employment experiences. Some participants held competitive, even executive, jobs and earned salaries well above the national average. However, the majority of adults with MMC in the study group reported an annual salary below the national average, which may be partly explained by the large proportion of participants in part-time employment. Participants who were employed reported significantly higher vocational and financial satisfaction. This highlights the importance of employment on adult life satisfaction and the need for additional resources to assist in finding employment which can accommodate individuals with cognitive disabilities and/or impairments in body structure and functions.

Participants with hydrocephalus and those lacking independent mobility experienced the greatest difficulty in living independently and forming romantic relationships. Several participants reported happy, healthy marriages and relationships, and this contributed significantly to life satisfaction. Overall, partner relationships were rated the least satisfying domain, indicating the need for additional attention to this aspect of adult life. Previous reports indicate that individuals with MMC have reduced social contact, which is clearly a barrier to developing romantic relationships.[27] Adolescents and adults with MMC should be encouraged to participate in social activities both with non-disabled peers as well as other individuals with MMC or similar disabilities. Involvement in online groups, which were not available to adults with MMC in the past, is not limited by physical disabilities and may increase opportunities for social contact.

While men with MMC are reported to have reduced fertility, similar frequencies of successful pregnancies and infertility were reported by men and women, though the number of participants who attempted pregnancy was low.[30,35] A reduced number of participants who conceived after the benefits of folic acid were known reported taking the therapeutic dose. Both women and men with MMC should be referred to a reproductive health specialist if they are considering pregnancy to discuss recurrence risks for NTDs, prenatal diagnostic options, and folic acid supplementation.

Women with MMC experienced increased frequencies of pregnancy complications, indicating a need for these

women to be monitored by high-risk obstetricians during pregnancy. Half of women in the study group delivered preterm, confirming previous reports of increased risk for preterm labor.[34] There is mention in the literature that women with MMC may benefit from or require C-section deliveries; however the majority of women in the study group were able to deliver vaginally.[31,34] In order to make informed decisions about pregnancy, health care providers should routinely incorporate discussions about sexuality and reproductive issues including alternatives such as gamete donation or surrogacy, from adolescence. Unfortunately, these topics are often overlooked leading to confusion and misconceptions.[19,21]

When compared to a reference group, adults with MMC in the study group were equally satisfied with life as whole, though several domains including partner relationships, finances and employment, were rated significantly lower. This suggests that while MMC does not affect overall life satisfaction for adults, actions are needed to increase satisfaction with specific aspects of adult life. Participants who lived with someone, including those residing in group homes, were significantly more satisfied with life as a whole than those participants living alone. While being self-sufficient is an important goal, living alone may contribute to decreased life satisfaction due to persistent social isolation. Isolated adults with MMC could consider seeking out roommates, perhaps by enlisting the help of local support group chapters.

There were several limitations in the present study. The twelve participants born before 1960 represent an especially fit group of individuals with MMC that may not translate to populations of adults born after 1960, when individuals with greater disabilities began to survive. However, multivariate analyses determined that the absence of hydrocephalus, rather than age alone, accounted for the generally positive outcome of these adults. Secondly, in some cases, a parent rather than the individual with MMC completed the questionnaire. Therefore, responses, especially in regards to life satisfaction, may not truly reflect those of the individual with MMC. However, a previous study examining quality of life in adolescents with MMC demonstrated that parent report closely mirrored self-report.[38] Additionally, participants were recruited through an NTD genetics study and therefore often had an additional family member with an NTD. This study group may represent a population of individuals with more available resources or involvement in the community and may not represent all individuals with MMC. Lastly, the study had a relatively small sample size consisting of 90 adult participants with MMC.

This is one of the few outcome and life satisfaction studies on adults with MMC. Results of this study have practical implications for providing information about long-term outcomes for individuals with MMC. This information may be valuable for parents in making pregnancy decisions or to aid in developing realistic goals for adolescents with MMC in their transition to adulthood.[25] This study also provides insight into areas of needed improvement in the care of adults with MMC. In particular, there is a clear need for information regarding reproductive issues for both men and women with MMC. Future research should focus on areas of reported difficulty and those domains with reduced life satisfaction to better understand the needs of adults with MMC.

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References

1. Detrait ER, George TM, Etchevers HC, Gilbert JR, Vekemans M, Speer MC. Human neural tube defects: developmental biology, epidemiology, and genetics. *Neurotoxicol Teratol*. 2005;27:515−524.
2. Dias MS, Partington M. Embryology of myelomeningocele and anencephaly. *Neurosurg Focus*. 2004;16:E1.
3. Bowman RM, McLone DG, Grant JA, Tomita T, Ito JA. Spina bifida outcome: a 25-year prospective. *Pediatr Neurosurg*. 2001;34:114−120.
4. 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.
5. McDonnell GV, McCann JP. Issues of medical management in adults with spina bifida. *Childs Nerv Syst*. 2000;16:222−227.
6. Oakeshott P, Hunt GM. Long-term outcome in open spina bifida. *Br J Gen Pract*. 2003;53:632−636.
7. Roach JW, Short BF, Saltzman HM. Adult consequences of spina bifida: a cohort study. *Clin Orthop Relat Res*. 2011;469:1246−1252.
8. Shin M, Besser LM, Siffel C, et al. Prevalence of spina bifida among children and adolescents in 10 regions in the United States. *Pediatrics*. 2010;126:274−279.
9. Sandler AD. Children with spina bifida: key clinical issues. *Pediatr Clin North Am*. 2010;57:879−892.
10. Barf HA, Verhoef M, Jennekens-Schinkel A, Post MW, Gooskens RH, Prevo AJ. Cognitive status of young adults with spina bifida. *Dev Med Child Neurol*. 2003;45:813−820.
11. Barf HA, Verhoef M, Post MW, et al. Educational career and predictors of type of education in young adults with spina bifida. *Int J Rehabil Res*. 2004;27:45−52.
12. Barf HA, Post MW, Verhoef M, Jennekens-Schinkel A, Gooskens RH, Prevo AJ. Life satisfaction of young adults with spina bifida. *Dev Med Child Neurol*. 2007;49:458−463.
13. Barf HA, Post MW, Verhoef M, Jennekens-Schinkel A, Gooskens RH, Prevo AJ. Restrictions in social participation of young adults with spina bifida. *Disabil Rehabil*. 2009;31:921−927.
14. Bellin MH, Dicianno BE, Levey E, et al. Interrelationships of sex, level of lesion, and transition outcomes among young adults with myelomeningocele. *Dev Med Child Neurol*. 2011;53:647−652.
15. Buffart LM, van den Berg-Emons RJ, van Meeteren J, Stam HJ, Roebroeck ME. Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele. *Dev Med Child Neurol*. 2009;51:886−894.
16. Laurence KM, Beresford A. Continence, friends, marriage and children in 51 adults with spina bifida. *Dev Med Child Neurol Suppl*; 1975:123−128.

17. van Mechelen MC, Verhoef M, van Asbeck FW, Post MW. Work participation among young adults with spina bifida in the Netherlands. *Dev Med Child Neurol*. 2008;50:772−777.
18. Rintoul NE, Sutton LN, Hubbard AM, et al. A new look at myelomeningoceles: functional level, vertebral level, shunting, and the implications for fetal intervention. *Pediatrics*. 2002;109:409−413.
19. Verhoef M, Barf HA, Vroege JA, et al. Sex education, relationships, and sexuality in young adults with spina bifida. *Arch Phys Med Rehabil*. 2005;86:979−987.
20. Hetherington R, Dennis M, Barnes M, Drake J, Gentili F. Functional outcome in young adults with spina bifida and hydrocephalus. *Childs Nerv Syst*. 2006;22:117−124.
21. Sawyer SM, Roberts KV. Sexual and reproductive health in young people with spina bifida. *Dev Med Child Neurol*. 1999;41:671−675.
22. Zukerman JM, Devine KA, Holmbeck GN. Adolescent predictors of emerging adulthood milestones in youth with spina bifida. *J Pediatr Psychol*. 2011;36:265−276.
23. Lemelle JL, Guillemin F, Aubert D, et al. Quality of life and continence in patients with spina bifida. *Qual Life Res*. 2006;15: 1481−1492.
24. Klingbeil H, Baer HR, Wilson PE. Aging with a disability. *Arch Phys Med Rehabil*. 2004;85:S68−S73.
25. Oakeshott P, Hunt GM, Poulton A, Reid F. Expectation of life and unexpected death in open spina bifida: a 40-year complete, nonselective, longitudinal cohort study. *Dev Med Child Neurol*. 2010;52:749−753.
26. Thibadeau JK, Alriksson-Schmidt AI, Zabel TA. The National Spina Bifida Program transition initiative: the people, the plan, and the process. *Pediatr Clin North Am*. 2010;57:903−910.
27. Liptak GS, Kennedy JA, Dosa NP. Youth with spina bifida and transitions: health and social participation in a nationally represented sample. *J Pediatr*. 2010;157:584−588.
28. Castree BJ, Walker JH. The young adult with spina bifida. *Br Med J (Clin Res Ed)*. 1981;283:1040−1042.
29. Magill-Evans J, Galambos N, Darrah J, Nickerson C. Predictors of employment for young adults with developmental motor disabilities. *Work*. 2008;31:433−442.
30. Decter RM, Furness PD 3rd, Nguyen TA, McGowan M, Laudermilch C, Telenko A. Reproductive understanding, sexual functioning and testosterone levels in men with spina bifida. *J Urol*. 1997;157:1466−1468.
31. Gatti C, Del Rossi C, Ferrari A, Casolari E, Casadio G, Scire G. Predictors of successful sexual partnering of adults with spina bifida. *J Urol*. 2009;182:1911−1916.
32. Carter CO, Evans K. Children of adult survivors with spina bifida cystica. *Lancet*. 1973;2:924−926.
33. Richmond D, Zaharievski I, Bond A. Management of pregnancy in mothers with spina bifida. *Eur J Obstet Gynecol Reprod Biol*. 1987;25:341−345.
34. Rietberg CC, Lindhout D. Adult patients with spina bifida cystica: genetic counselling, pregnancy and delivery. *Eur J Obstet Gynecol Reprod Biol*. 1993;52:63−70.
35. Woodhouse CR. Myelomeningocele in young adults. *BJU Int*. 2005;95:223−230.
36. Deak KL, Siegel DG, George TM, et al. Further evidence for a maternal genetic effect and a sex-influenced effect contributing to risk for human neural tube defects. *Birth Defects Res A Clin Mol Teratol*. 2008;82:662−669.
37. Prevention of neural tube defects: results of the Medical Research Council Vitamin Study. MRC Vitamin Study Research Group. *Lancet*. 1991;338:131−137.
38. Sawin KJ, Brei TJ, Buran CF, Fastenau PS. Factors associated with quality of life in adolescents with spina bifida. *J Holist Nurs*. 2002;20:279−304.
39. Sawin KJ, Bellin MH. Quality of life in individuals with spina bifida: a research update. *Dev Disabil Res Rev*. 2010;16:47−59.
40. Verhoef M, Post MW, Barf HA, van Asbeck FW, Gooskens RH, Prevo AJ. Perceived health in young adults with spina bifida. *Dev Med Child Neurol*. 2007;49:192−197.
41. Fugl-Meyer AR, Melin R, Fugl-Meyer KS. Life satisfaction in 18- to 64-year-old Swedes: in relation to gender, age, partner and immigrant status. *J Rehabil Med*. 2002;34:239−246.
42. United States Department of Labor, Bureau of Labor Statistics. *Occupational Classification System Manual (OCSM)*, http://www.bls.gov/ocsm/commain.htm; 2002.
43. United States Census Bureau. *Income, Poverty, and Health Insurance Coverage in the United States: 2010*, http://www.census.gov/prod/2011pubs/p60-239.pdf; 2011.
44. Goodwin P, McGill B, Chandra A. Who marries and when? Age at first marriage in the United States: 2002. *NCHS Data Brief*; 2009:1−8.
45. Adzick NS, Thom EA, Spong CY, et al. A randomized trial of prenatal versus postnatal repair of myelomeningocele. *N Engl J Med*. 2011;364:993−1004.
46. United States Census Bureau. *Educational Attainment in the United States: 2010*, http://www.census.gov/hhes/socdemo/education/; 2011.
47. English LH, Barnes MA, Taylor HB, Landry SH. Mathematical development in spina bifida. *Dev Disabil Res Rev*. 2009;15:28−34.