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Original Research Article

Cross-Lag Model of Medical Responsibility and Skills in Youth With Spina Bifida

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Abstract

Objective This study examined bidirectional associations between mother- and father-reported medical responsibility and medical skill mastery in youth with spina bifida (SB). **Methods** Participants were 140 youth with SB and their parents who participated in three waves of a longitudinal study across four years (ages 8–15 years at Time 1). Mother- and father-report of both medical responsibility and medical skill mastery were used, and age and estimated intelligence quotient were included as covariates, in cross-lagged models. **Results** The cross-lagged model provided evidence for significant bidirectional associations between mother-reported medical responsibility and skill mastery across time (root mean square error of approximation=0.09, comparative fix index=0.97). These paths showed that higher levels of child responsibility predicted an increase in skill mastery and that higher levels of mastery predicted an increase in child responsibility across time. Moreover, based on mother-report, sharing of responsibility had stronger effects on increases in skill mastery (Time 1 to Time 2 β=.25, Time 2 to Time 3 β=.27) than skill mastery had on increases in child responsibility (Time 1 to Time 2 β=.08, Time 2 to Time 3 β=.07). The only significant cross-lagged path for father-report was from Time 1 skill mastery to Time 2 responsibility (β=.34). **Conclusions** Mothers perceive a bidirectional relationship between responsibility and skill mastery across time, whereas fathers appear to mainly consider how skills might affect a subsequent increase in responsibility sharing. Thus, it is important to consider both parents' perspectives when working to increase medical autonomy in youth with SB.

Key words: spina bifida; longitudinal; medical responsibility; medical skill mastery; medical autonomy.

Introduction

Spina bifida (SB) is a relatively common congenital birth defect that affects ~3 out of every 10,000 births in the United States (National Birth Defects Prevention Network, 2010). Youth with SB require multifaceted medical care, including regular clinic visits, daily medication, dietary restrictions, catheterization, and bowel management programs (Copp et al., 2015). Such medical treatments are complex, intrusive, and time consuming, adding burden on families and youth who are already navigating the challenges of childhood and adolescence. In fact, with innovations in these treatments, more responsibility is falling on youth and families to manage their own medical regimen, which has given rise to research on pediatric self-management (Grey et al., 2015; Modi et al., 2012; Reed-Knight et al., 2014; Ryan & Sawin, 2009; Sawin, 2017).

Typically, youth with SB begin learning medical care skills in early childhood, but they continue to be dependent on adults for guidance as they begin to take responsibility for various components of their medical regimen (Psihogios et al., 2015; Quittner et al., 2008). There are multiple factors that influence the degree and quality of self-management, including nonmodifiable individual level variables (e.g., age, intelligence

quotient [IQ]), modifiable individual variables (e.g., mastery of regimen-related skills), and family-level variables (e.g., allocation of responsibility sharing; Modi et al., 2012; Reed-Knight et al., 2014). Existing theories of self-management suggest that decisions to transfer medical responsibility to children and adolescents are often based on their assessment of the youth's level of condition knowledge, mastery of medical skills, and self-efficacy (i.e., their belief that they are able to successfully execute necessary medically relevant behaviors) after taking into account nonmodifiable variables (Modi et al., 2012; Reed-Knight et al., 2014; Wiebe et al., 2014).

Unfortunately, we know little about parents' perceptions of how medical responsibility sharing and skill mastery influence each other across time. For example, does skill mastery account for most of the change in responsibility sharing across time? Do parents believe that gradually increasing levels of responsibility (regardless of prior levels of mastery) will lead to increasing levels of skill mastery as they learn from repeated, supervised attempts at self-management (i.e., should parents assume that children will learn from their own mistakes?)? Or, most likely, do parents consider both current responsibility sharing and skill mastery to determine how medical regimen tasks will be distributed in the future? In this study, we sought to examine the interplay over time between two components of what has been referred to as "medical autonomy," namely, medical responsibility and medical skill mastery (Psihogios et al., 2015; Wysocki et al., 1996a).

Until now, theorizing about and studies focused on constructs related to medical autonomy have tended to focus only on predictors of subsequent levels of youth responsibility (Holmes et al., 2006; Psihogios et al., 2015; Reed-Knight et al., 2014; Wiebe et al., 2014). The Pediatric Self-Management Theory, however, suggests a more nuanced approach to medical autonomy, proposing that it consists of the interaction of medical responsibility sharing and skill mastery across time (Modi et al., 2012). Responsibility sharing and skill mastery are distinct, but interrelated constructs, and are the primary factors shown to be related to the transfer of health care responsibility (Pai & Ostendorf, 2011; Reed-Knight et al., 2014). Despite responsibility sharing and skill mastery being theorized as two constructs that are part of an overarching medical autonomy factor (Modi et al., 2012), much less is known about how these variables influence each other across time. Bidirectional effects illuminate, for both families and clinicians, how parents perceive that these variables influence each other over time and whether one may be more influential than the other in increasing pediatric self-management skills. Reed-Knight et al. (2014) explain that there are multiple "errors" that can occur in this process (e.g., giving responsibility too early when adolescent is not prepared, giving responsibility too late and denying the adolescent adequate learning opportunities). By examining these constructs bidirectionally, we gain a better understanding of how parents perceive that responsibility sharing and skill mastery influence each other, which allows clinicians and families to make more informed discussions regarding medical autonomy.

The purpose of this study was to use cross-lag models to examine bidirectional associations between youth management of medical responsibilities and their mastery of medical care skills across four years in families of youth with SB. Longitudinal research is ideal for studying these processes because it allows one to tease apart bidirectional influences between two or more variables. In other words, with such analyses, we will be able to assess how much each of these variables influence each other across time. Additionally, cross-lag analyses enable one to examine the differential strength of the pathways as the variables of interest interact with each other across time so that we can understand how much skill mastery and responsibility sharing influence each other, while accounting for prior report on those variables. As cross-lag models also account for prior time points, such models are able to take various reporter biases into account when reporting estimates.

Past findings support that, with age, youth with chronic health conditions gain more medically related responsibility and skill mastery (Psihogios et al., 2015; Reed-Knight et al., 2014; Wiebe et al., 2014). Recent work with youth who have SB has shown that there are two distinct trajectories for how the management of medical responsibilities changes across time (Kayle et al., 2020). The first trajectory group includes two-thirds of youth with SB who are granted increasing levels of medical responsibility during late childhood and adolescence and start at higher levels of responsibility, whereas the other one-third of youth with SB start at a significantly lower level of responsibility with a much less rapid increase in responsibility (Kayle et al., 2020). With respect to specific medical tasks, most tasks remain at least partially within parental control, with the exception of catheterization (i.e., parents report that 67–80% of teens are primarily responsible for catheterization). In fact, only 38–56% of 16- or 17-year-old adolescents with SB take *responsibility* for their bowel programs, skin checks, and exercise programs according to their parents (Psihogios et al., 2015). Despite this, parents report that their children have *mastered* each of these skills between 69% and 90% of the time (Psihogios et al., 2015). This discrepancy between responsibility sharing and skill

mastery may be due to continuing parental supervision, which appears to play a role in the transfer of medical responsibility (Driscoll et al., 2020). In addition, across a host of medical conditions (e.g., type 1 diabetes), adherence often dips during adolescence as youth gain more control over their medical regimen, possibly leading parents to increase their level of support even when adolescents appear to be capable of completing the medical tasks (Psihogios et al., 2015; Reed-Knight et al., 2014).

At the cross-sectional level, higher levels of medical responsibility are moderately to strongly associated with level of skill mastery in youth and young adults with SB and mother- and father-report of both constructs are strongly associated (Psihogios et al., 2015; Smith & Holmbeck, 2021). Unfortunately, such findings do not permit conclusions about how each of these self-management constructs affect each other over time. One study used longitudinal regression analyses to examine whether skill mastery at Time 1 predicted responsibility sharing at Time 2 (Psihogios et al., 2015). Father-reported skills predicted later father-reported responsibility ($\beta = .54$, $p < .05$), but this was not true for mother-report of skills and responsibility ($\beta = .34$, $p = .13$). There are several limitations to these analyses: only two time points were examined, list-wise deletion was used to handle missing data, and the association between skill mastery and subsequent responsibility was examined without assessing the bidirectional relationship. This study will address these limitations by being the first to use cross-lagged models to examine bidirectional longitudinal associations between medical responsibility management and regimen skill mastery in youth with SB, based on mother and father report, using three time points across four years. Separate models for mother- and father-report were created due to differences between parents in their involvement with and in their perceptions of youth responsibility and mastery (Brekke et al., 2017; Psihogios et al., 2015). This study builds upon prior research that shows that both child responsibility and skill mastery increase across time and expands upon these findings by including both constructs within the same analytic model. It was hypothesized that higher rates of skill mastery would predict an increase in child responsibility and also that higher levels of child responsibility would predict an increase in skill mastery. We also compared the strength of the cross-lag pathways from skill mastery to responsibility versus responsibility to skill mastery.

Methods

Participants

Participants were part of a larger, longitudinal study at Loyola University Chicago that is examining family, psychosocial, and neurocognitive functioning among children with SB (e.g., Stern et al., 2020). This report used data regarding health care behaviors from Time 1 (ages 8–15), Time 2 (ages 10–17), and Time 3 (ages 12–19). Families of children with SB were recruited from four hospitals and a statewide SB association in the Midwest. Inclusion criteria consisted of: (a) diagnosis of SB (types included myelomeningocele, lipomeningocele, or myelocystocele); (b) age 8–15 years at Time 1; (c) ability to speak or read English or Spanish; (d) involvement of at least one primary caregiver; and (e) residence within 300 miles of the lab to allow for home-based data collections. Of the original 246 families who met eligibility criteria, 163 families agreed to participate, but 21 of those families could not be contacted or later declined, and two families eventually did not meet inclusion criteria (i.e., one child was too young and one had a milder form of SB).

The final sample of participants included 140 families of children with SB at Time 1 (53.6% female; M_{age} =11.43). Of these children, 52.9% identified as Caucasian, 27.9% were Hispanic/Latinx, 13.6% were African American, 1.4% were Asian, and 4.3% identified as bi-racial. The average Hollingshead Four Factor Index for the sample was ~39.12 (SD = 16.09), suggesting a generally middle-class sample with some variability. Children of families who declined participation did not differ from those who participated with respect to type of SB (e.g., myelomeningocele or other), χ^2 (1) = 0.0002, $p > .05$, shunt status, χ^2 (1) = .003, $p > .05$, or occurrence/nonoccurrence of shunt infections, χ^2 (1) = 1.08, $p > .05$. At Time 2 (T2; ages 10–17) and Time 3 (T3; ages 12–19), 110 and 104 youth with SB participated, respectively. See Table I for more details about the sample. Importantly, beginning at T3, parents of participants who had turned 18 (roughly 25% of the sample at T3) no longer participated in the home visit data collections. Thus, data were not collected from parents for the medical autonomy variables if their child was over 18 at T3. This affected 28 participants for the mother-report model and 26 participants for the father-reported model. These families were still included in the analyses at Time 1 and Time 2 and self-report did not replace parent-report in these models. Sample size at Time 1 for mother-report was 118, Time 2 ($N = 99$), and Time 3 ($N = 96$) and for father-report $N = 95$, $N = 76$, and $N = 51$ respectively. Attrition analyses indicated that families who did not participate at T2 or T3 did not significantly differ from those who did with respect to gender, estimated IQ, age, lesion level, medical responsibility, and medical skill mastery.

Child medical information regarding physical health status was gathered from medical charts (medical chart release was obtained during home visit) and

Table I. *Demographic and Condition-Specific Characteristics at Time 1*

Demographics	Youth with SB ($N = 140$)
Age, *M* (SD)	
T1	11.43 (2.46)
T2	13.37 (2.43)
T3	15.36 (2.43)
Sex	
Male	65 (46.4%)
Female	75 (53.6%)
Race/ethnicity	
Caucasian	74 (52.9%)
Hispanic/Latino	39 (27.9%)
African American	19 (13.6%)
Asian	2 (1.4%)
Bi-racial	6 (4.3%)
SB type	
Myelomeningocele	122 (87.1%)
Other	18 (12.9%)
Lesion level	
Thoracic	23 (16.4%)
Lumbar	69 (49.3%)
Sacral	41 (29.3%)
Unknown/not reported	7 (5.0%)
Shunt status: present	109 (77.9%)
FSIQ at T1, *M* (SD)	85.75 (19.54)
MR T1 SOSBMR	1.76 (0.17)
MR T2 SOSBMR	1.96 (0.20)
MR T3 SOSBMR	2.07 (0.18)
MR T1 SBIS	0.67 (0.07)
MR T2 SBIS	0.75 (0.06)
MR T3 SBIS	0.82 (0.05)
FR T1 SOSBMR	1.69 (0.17)
FR T2 SOSBMR	1.86 (0.22)
FR T3 SOSBMR	2.02 (0.18)
FR T1 SBIS	0.67 (0.08)
FR T2 SBIS	0.72 (0.07)
FR T3 SBIS	0.81 (0.05)

Note. SB characteristics were reported by parents and confirmed with medical chart review. SB = spina bifida; M = mean; SD = standard deviation; T1 = Time 1; T2 = Time 2; T3 = Time 3; FSIQ = Estimated Full Scale Intelligence Quotient, MR = mother-report, FR = father-report.

questionnaire data. Of the 140 participants, medical chart data indicated the following diagnosis rates: 87.1% myelomeningocele and 12.9% other. Additionally, over half of the children had spinal lesions located in the lumbrosacral or lumbar spinal regions (49.3%), 29.3% had lesions in the sacral region, and 16.4% had lesions in the thoracic region. Medical chart data showed that 77.9% of the children had a shunt, and mother questionnaire data indicated the average number of shunt surgeries at Time 1 was 3.14 (SD = 5.07). According to parent-reported questionnaire data, 81.1% of the children used braces to ambulate and 61.4% used a wheelchair (as some children used both methods of ambulation). Similar to past studies (e.g., Wills et al., 1990), youth with SB demonstrated a low average IQ (*M* Full Scale estimated IQ = 85.75, SD = 19.54).

Procedure

This study was approved by university and hospital Institutional Review Boards. For full study procedures, please see (Stern et al., 2020). Time points (i.e., T1–3) occurred ~2 years apart. At T1, data were collected across two in-home assessment sessions conducted by two trained research assistants. At T2 and T3, data were collected during single in-home assessment sessions. Informed child assent and parental consent were obtained prior to all data collections. Trained undergraduate and graduate student research assistants collected data during scheduled home visits that lasted ~3 hr. Families who completed all parts of the study received monetary compensation ($150 for families) and gifts (e.g., t-shirts and pens). Youth with SB and their parents independently completed questionnaires in separate rooms. Research assistants read questionnaires out loud to participants when requested or when reading difficulties were observed or described by youth or parents.

Measures

Demographics and SB Characteristics

Parents of children with SB completed a questionnaire detailing the child's demographic information (e.g., age, race/ethnicity, etc.). SB characteristics (e.g., type of SB, lesion level) were reported by parents and confirmed with medical chart review. Full scale estimated IQ was determined at T1 for each participant based on their performance on two subtests from the Weschler Abbreviated Scale of Intelligence (Zhu, 1999).

Sharing of Medical Responsibilities

The Sharing of SB Management Responsibilities Scale (SOSBMR), adapted from the Diabetes Family Responsibility Questionnaire (Anderson, et al., 1990), was used to assess parent perceptions of the division of SB responsibilities within the family. The SOSBMR consists of 34 items that describe SB and health-related issues relevant to children with SB (e.g., remembering to catheterize regularly, every 2–4 hr). Parents rated who was primarily responsible for each task (e.g., parent, child, shared, or not applicable). The SOSBMR has demonstrated adequate internal consistency and concurrent validity in youth and young adults with SB from the same data set (Psihogios et al., 2015; Smith & Holmbeck, 2021). Smith and Holmbeck (2021) found that the one-factor solution (all 34 items loading on a single SOSBMR factor) had good fit (mother-report root mean square error of approximation [RMSEA] = 0.06, comparative fix index [CFI] = 0.94, Tucker–Lewis index [TLI] = 0.93, father-report RMSEA = 0.08, CFI = 0.95, TLI = 0.95) and was moderately associated with other measures of medical autonomy (e.g., mastery of

medical skills), thus demonstrating satisfactory construct validity (Psihogios et al., 2015). The SOSBMR yielded satisfactory reliability for mother-report (reliability coefficient = .95) and adequate reliability for father-report (.68).

Medical Skill Mastery

The Spina Bifida Independence Survey (SBIS), adapted from the Diabetes Independence Survey (Wysocki et al., 1996a), was used to measure parent's evaluation of the child's attainment of SB skills. The SBIS is composed of 50 SB-specific skill questions to which participants respond "yes," "no," "not sure," or "not applicable" for each item that assessed whether the child was able to correctly perform each skill independently (e.g., "Does your child move in and out of your/their wheelchair at home?," "Does your child do each catheterization step correctly?"). Recent work has shown excellent fit for the one-factor model for both mother- (RMSEA = 0.04, CFI = 0.96, and TLI = 0.96) and father-report (RMSEA = 0.03, CFI = 0.98, and TLI = 0.98; Smith & Holmbeck, 2021). The SBIS has good reliability (mother-report reliability coefficient = .96, father-report = .96) and has been found to be positively associated with adherence, management of responsibilities, and age (Psihogios et al., 2015).

Data Analysis

To examine bidirectional associations between parent-reported medical responsibility management and skill mastery, cross-lagged panel models were estimated using M*plus* Version 8 (Muthén & Muthén, 1998–2017). Mother- and father-report were examined in separate models and estimated IQ, lesion level, and age were examined as covariates. Medical autonomy variables at nonsequential time points (i.e., SOSBMR at T1 and T3, SBIS at T1 and T3) were allowed to covary and were included in the model to partially account for shared method variance. Additionally, each of the medical autonomy variables was allowed to covary at the same time point (e.g., SOSBMR with SBIS at T1). A bidirectional model was tested for both mother- and father-report in which medical responsibility and medical skill mastery were regressed on the variables from the preceding time point. As prior work has suggested that age may moderate the trajectory of responsibility and skill mastery, we examined whether age at T1 moderated the cross-lagged coefficients. Age was broken into 8–11 and 12–15 to examine whether there were different trajectories for younger and older age groups.

For all analyses, full information maximum likelihood was used to address missing data (Enders & Bandalos, 2001). All observed information is used to estimate parameters with this method. To be able to compare effect sizes, standardized betas are reported. Goodness of fit for the estimated models were examined by various fit indices including the CFI (ideal study criterion $\geq$ 0.95), TLI (ideal study criterion $\geq$ 0.95), RMSEA (ideal study criterion $\leq$ 0.05), and standarized root mean squared residual (SRMR, ideal study criterion $\leq$ 0.10; Hu & Bentler, 1999; Kline, 2011). A change in CFI (ΔCFI) above 0.01 was used to determine whether a more constrained model resulted in a significant deterioration in model fit over the less restrictive model (Cheung & Rensvold, 2002). The overall global model fit (i.e., highest CFI and TLI indices; lowest RMSEA indices) was also assessed to determine best fitting models.

Results

For both reporters, the best fitting model included covarying age at T1 and estimated IQ covarying at all three time points. Lesion level was not significant with SOSBMR and SBIS and worsened model fit, so was dropped as a covariate.

Mother-Reported Medical Autonomy Cross-Lagged Model

SOSBMR (i.e., responsibility) at T1 and T3 and SBIS (i.e, skill mastery) at T1 and T3 were allowed to covary. In addition, SOSBMR at T1 and SBIS at T1, SOSBMR at T2 and SBIS at T2, and SOSBMR at T3 and SBIS at T3 were allowed to covary. The bidirectional model with medical responsibility and mastery of skills demonstrated adequate fit across all indices (χ^2 = 17.28, p = .03, RMSEA = 0.09, CFI = 0.97, TLI = 0.90, and SRMR = 0.06). In this model, evidence was found for bidirectional effects between medical responsibility and skill mastery, with greater child responsibility predicting higher levels of skill mastery (T1–2, β = .25, p < .001; T2–3, β = .27, p < .001) and higher levels of skill mastery predicting more child medical responsibility (T1–2, β = .08, p < .001; T2–3, β = .07, p < .001) across 4 years. Importantly, pathways from responsibility to skills were stronger than the pathways from skills to responsibility. Additionally, as expected, youth that were higher on medical responsibility at the preceding time point were higher on medical responsibility at later time points. The same was true for skill mastery over time. Specifically, medical responsibility at T1 predicted responsibility at T2 (β = .79, p < .001) and T2 medical responsibility predicted T3 responsibility (β = .44, p < .001). Similarly, T1 skill mastery predicted T2 skills (β = .59, p < .001) and T2 skills predicted T3 skills (β = .32, p = .037). See Figure 1 for the full mother-report model.

Father-Reported Medical Autonomy Cross-Lagged Model

This model was run in the same manner as the model based on maternal report. The bidirectional model with medical responsibility and skill mastery demonstrated good fit across all indices ($\chi^2 = 7.95$, $p = .16$, RMSEA = 0.06, CFI = 0.99, TLI = 0.93, and SRMR = 0.05). In this model, only one cross-lagged path was significant, with T1 skill mastery predicting T2 responsibility management ($\beta = .34$, $p = .004$). As was found with the mother-report model, all sequential effects within the same medical autonomy variable were significant. That is, having higher levels of medical responsibility at T1 predicted higher child medical responsibility at T2 ($\beta = .46$, $p < .001$) and more child responsibility at T2 predicted higher levels of responsibility at T3 ($\beta = .86$, $p < .001$). Similarly, having higher levels of skill mastery at T1 predicted higher levels of mastery at T2 ($\beta = .77$, $p < .001$) and having higher levels of skill mastery at T2 predicted higher levels of skill mastery at T3 ($\beta = .40$, $p = .024$). See Figure 2 for the full father-report model.

Age as Moderator

Using the final mother- and father-report models, age at T1 was included as a moderator of each of the cross-lagged paths. For both reporters, including age as a moderator significantly decreased model fit and the moderation was not significant. For mother-report, age did not significantly moderate the path from T1 responsibility to T2 skill mastery ($\beta = -.06$, $p = .40$) nor did it moderate the path from T1 skill mastery to T2 responsibility ($\beta = -.02$, $p = .33$). Age did not moderate the path from T2 responsibility to T3 skill mastery ($\beta = -.02$, $p = .67$) nor T2 skill mastery to T3 responsibility ($\beta = -.08$, $p = .59$). For father-report, age did not significantly moderate T1 responsibility to T2 skill mastery ($\beta = .03$, $p = .20$), T2 responsibility to T3 skill mastery ($\beta = -.01$, $p = .77$), T1 skill mastery to T2 responsibility ($\beta = .06$, $p = .63$), nor T2 skill mastery to T3 responsibility ($\beta = .10$, $p = .71$). Thus, the final models were kept without age as a moderator.

Discussion

The purpose of this study was to examine bidirectional longitudinal associations between medical responsibility and medical skill mastery in youth with SB. Consistent with the hypotheses, the results of this study suggested that higher levels of mother-reported child skill mastery predicted an increase in responsibility for medical care and vice versa. Interestingly, even though the pathways were significant in both directions for mother-report, the strongest cross-lagged effects were from medical responsibility to skill mastery rather than skill mastery to responsibility. However, the cross-lag effect sizes were small regardless of the direction of the association. For father-report, only T1 skill mastery predicted T2 child responsibility management. Overall, the results revealed that mothers and fathers differ in how they consider sharing responsibility. For mothers, there is a bidirectional, longitudinal association, where mothers appear to take into account both skill mastery and responsibility over time when considering who will take charge of the medical regimen. For fathers, it may be that they expect some skill mastery before increased medical responsibility, but the connection between these two constructs appears to dissipate over time. In addition, fathers perceived that having higher levels of both skill mastery and child responsibility of medical tasks at an earlier time point would predict having higher levels of both skill mastery and responsibility at subsequent time points, respectively.

Although the mother-report model revealed significant cross-lagged pathways in both directions for responsibility and skill mastery, the effects were stronger for the pathways from child responsibility to skill mastery. This finding suggests that mothers perceive that sharing responsibility of medical tasks has more influence over skill mastery than skill mastery has on their perception of increases in responsibility sharing. At first glance, this finding was somewhat unexpected, as theory would suggest that parents would perceive skill mastery as being influential in the process by which medical responsibility is transferred from parent to child (e.g., Reed-Knight et al., 2014). Moreover, the result also appears to be contrary to prior findings that suggest that mothers view children with SB as vulnerable and, consequently, mothers appear to continue to maintain responsibility for medical tasks even when their children have already developed the necessary skills (Driscoll et al., 2020; Psihogios et al., 2015). Importantly, these results suggest that mothers appear to believe that both skill mastery and responsibility sharing influence each other, but that responsibility sharing has more influence on skill mastery than vice versa. In this way, when children obtain responsibility, they are effectively given time to learn and practice their skills, presumably leading to more mastery. Learning theories suggest that higher levels of deliberate practice are necessary to attain mastery (Ackerman, 1987; Ericsson et al., 1993; Macnamara et al., 2014). To reach mastery of a skill, youth need to practice and learn from their mistakes (Ackerman, 1987). For example, when learning to ride a bike, parents often need to physically and mentally "let go" to allow their child to learn. Similarly, when mothers give their children and adolescents a somewhat higher level of responsibility, youth can begin to gain mastery in these skill areas. Thus, these findings suggest that helping mothers to gradually increase their granting of medical responsibility (with supervision) will likely

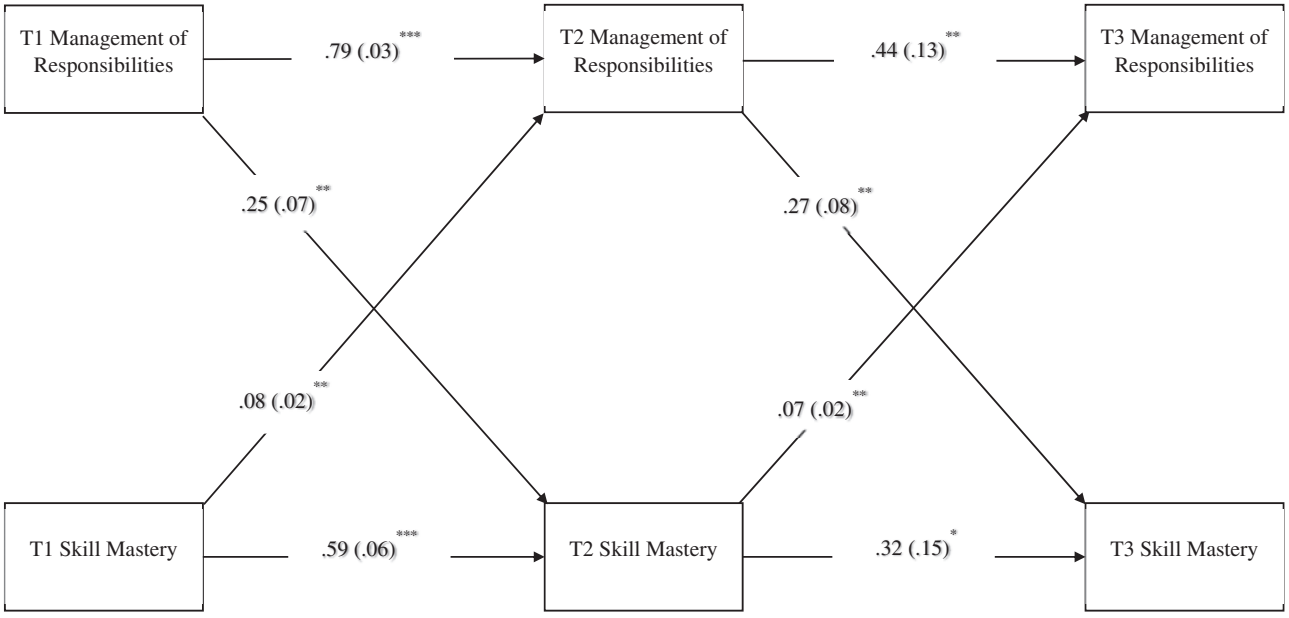

Figure 1. Cross-lagged effects model for mother-reported management of spina bifida responsibilities and medical skill mastery controlling for intelligence quotient (IQ) and age. *Note.* Standardized estimates are reported with standard errors in parentheses. Residuals and covariances are not shown for readability. Covariances between responsibility and skill mastery ranged from ($\beta = .12$, $p = .492$) to ($\beta = .53$, $p < .001$). Age was significantly associated with Sharing of SB Management Responsibilities Scale (SOSBMR; $\beta = .38$, $p < .001$) and Spina Bifida Independence Survey (SBIS; $\beta = .22$, $p = .024$) and associations for estimated IQ ranged from $\beta = .24$, $p = .030$ to $\beta = .37$, $p < .001$ for SOSBMR and $\beta = .001$, $p = .994$ to $\beta = .162$, $p = .068$ for SBIS. Solid lines show significant paths. *$p < .05$, **$p < .01$, ***$p < .001$.

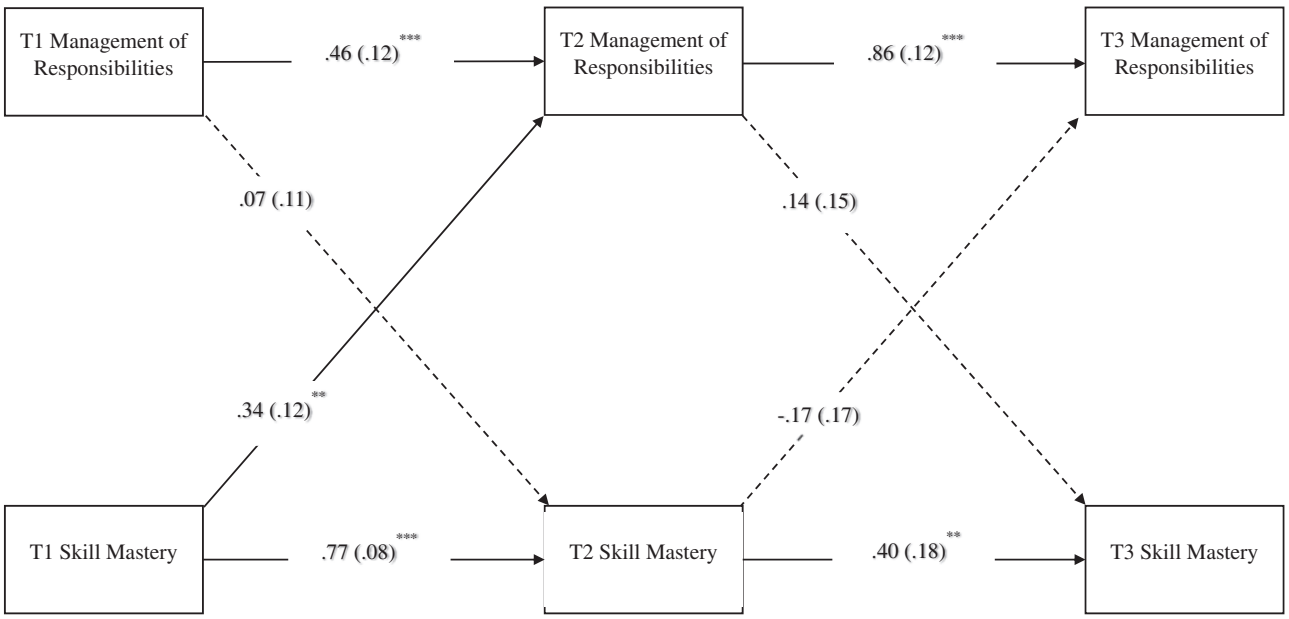

Figure 2. Cross-lagged effects model for father-reported management of spina bifida responsibilities and medical skill mastery controlling for intelligence quotient (IQ) and age. *Note.* Residuals and covariances are not shown for readability. Covariances between responsibility and skill mastery ranged from ($\beta = .20$, $p = .325$) to ($\beta = .64$, $p < .001$). Age was significantly associated with Sharing of SB Management Responsibilities Scale (SOSBMR; $\beta = .24$, $p = .022$) and Spina Bifida Independence Survey (SBIS; $\beta = .28$, $p = .003$) and associations for estimated IQ ranged from $\beta = .02$, $p = .916$ to $\beta = .39$, $p < .001$ for SOSBMR and $\beta = .02$, $p = .907$ to $\beta = .43$, $p < .001$ for SBIS. Solid lines show significant paths, dotted lines show nonsignificant paths. **$p < .01$, ***$p < .001$.

lead to a subsequent increase in skill mastery in their children.

Although the effects were stronger for the pathways from medical responsibility to skill mastery, these results also show that higher levels of mother-reported

skill mastery are associated with a subsequent increase in maternal perception of responsibility sharing, which is consistent with broader self-management theories (e.g., Reed-Knight et al., 2014; Wysocki et al., 1996b; Wiebe et al., 2014). Importantly, for both mothers and fathers, when parents perceived higher levels of medical responsibility or skill mastery at T1, that was associated with higher levels of the same construct at T2 (and the same was true for T2–3). This was only partially accounted for by the bidirectional effects and, as is typical when considering homotypic continuity (construct predicting same construct; Lahey et al., 2004), having higher levels of responsibility sharing predicted higher levels in later responsibility sharing and higher levels of skill mastery predicted higher levels in later skill mastery according to both parents. For fathers, higher levels of skill mastery were associated with an increase in child medical responsibility, but bidirectional effects were not found. This finding suggests that, unlike mothers, fathers perceive skill mastery as influencing responsibility sharing, but not vice versa. Fathers may believe skill mastery must come before increases in responsibility or that they perceive skill mastery and responsibility as being less connected than mothers across development. After Time 2, fathers continued to perceive that higher levels of T2 responsibility or skill mastery predicted higher levels of T3 responsibility or skill mastery respectively but that, over time, these variables were not perceived to be connected. It is also possible, but not tested in this study, that more responsibility may fall on mothers to make decisions regarding whether or not their child takes charge of medical tasks, which has been found in previous work (Brekke et al., 2017).

Interventions will need to address how mothers appear to attend to both responsibility and skill development simultaneously, whereas fathers appear to perceive that skill mastery influences shared medical responsibility. These results suggest that when working clinically with fathers of youth with SB, there may be two areas upon which to focus. First, fathers may benefit from increased psychoeducation regarding the interconnected relationship between skill mastery and responsibility. Second, clinicians could focus on teaching fathers how to increase their youth with SB's skill mastery, which in turn will lead to responsibility sharing. For mothers, who perceive responsibility sharing and skill mastery to be interrelated over time, providers can elicit discussions focused on the interconnectedness of these two self-management variables and use this understanding as a tool to help increase youth medical autonomy (if that is the goal of the clinician and the family). It is important to note, however, that clinicians should assess the levels of these perceptions in both parents regardless of gender and in the youth with SB to ascertain how each individual in the family unit assesses pediatric self-management of medical tasks. When discussing increasing youth medical autonomy with parents of youth with SB, it is important to assess not only whether parents perceive youth with SB as having the necessary skills, but also whether parents are actually sharing responsibilities with their child or adolescent with SB. If clinicians see a discrepancy between skill mastery and responsibility sharing, it may be beneficial to use motivational interviewing strategies with the family to understand the barriers to sharing of SB-related responsibilities (Schaefer & Kavookjian, 2017). Importantly, parents may benefit from receiving motivational interviewing, particularly when parental behavior change is expected (e.g., parents being willing to increase youth medical autonomy; Bean et al., 2014; Kitzmann et al., 2010). This study examined how parents perceive responsibility sharing and skill mastery, but there are multiple additional aspects to incorporate into future modeling, including youth motivation, self-efficacy, adherence, and disease knowledge (Modi et al., 2012). Moreover, it will be just as important to assess the perspective of youth with SB and ascertain whether a teen is motivated and ready to complete medical tasks on their own. For many youth with chronic illnesses, motivational interviewing helps increase adherence and self-management and can help lead to long-term positive health outcomes (Schaefer & Kavookjian, 2017).

This study had several strengths, including the use of longitudinal and multi-reporter data across three time points and the examination of bidirectional associations between two medical autonomy constructs. There are several limitations to this study, however, that should be addressed in future work. The primary objective of this study was to assess how mothers and fathers perceived responsibility sharing and medical skill mastery were interrelated across time. Importantly, these findings do not address which pathway is most *adaptive*. That is, these results only provide information about what is happening in families with youth with SB, not whether or not either of these pathways are associated with important outcomes, such as adherence and health complications.

There are also sample size limitations, which may be part of the reason that cross-lagged effects from T2 to T3 were not detected in the father-report model. Although the use of longitudinal data is indeed a strength, our use of longer time intervals (i.e., 2 years between sampling) may fail to accurately portray the interplay between increases in medical responsibility and increases in skill mastery. It will be important to gather information across smaller time intervals to more fully understand how the transfer of responsibility and skill mastery develop and influence each other over time. As there are many skill areas for youth with SB to manage, it is also important for future research

to examine the bidirectional associations of responsibility and skill acquisition for specific tasks (e.g., catherization, bowel management programs). It is possible that increase in a specific task (e.g., increased skill and responsibility in catheterization) would also increase skill mastery and/or responsibility in another medical task (e.g., bowel management programs). Additionally, this study specifically examined parent-report of medical autonomy, as parents often are the main decision makers when transferring medical responsibility; however, future work would benefit from gaining the perspective of youth on how they view their own levels of medical responsibility and skill mastery. Previous work has shown differences between parent and child reports of responsibility and skill mastery, with youth believing they have higher rates of responsibility and skill mastery than their parents report (Psihogios & Holmbeck, 2013; Psihogios et al., 2015). As adolescents get older, their autonomy tends to increase, as does their role in the self-management of medical tasks (Modi et al., 2012; Psihogios et al., 2015). In addition, although covariances were estimated in the models to partially account for shared method variance, it is important to note that cross-rater models were not examined. Relatedly, although cross-lag models account for prior reports on the variables of interest, it is possible there is still some confirmatory bias when only using parent-report.

For youth with SB, there is a bidirectional association between management of medical responsibilities and skill mastery from the mother's perspective. Mothers perceive responsibility sharing as influential for skill mastery and vice versa, whereas fathers perceive a positive relationship between their children's level of responsibility and skill mastery in only one direction (i.e., skill mastery leading to responsibility), which also dissipated over time. These findings help to further illuminate how parents perceive medical responsibilities to be transferred from parents to youth, a process which may serve as a developmental precursor to the successful transition to adult health care, gaining employment, and seeking higher education (Friedman et al., 2009; Warschausky et al., 2017). Importantly, these findings have implications for intervention, as clinicians and health care workers strive to help families increase adherence and medical autonomy for youth with SB. Given that parents often grapple with balancing the independence versus dependence needs of their children with SB (Sawin et al., 2003), they will likely benefit from interventions that target both the sharing of medical responsibilities and medical skill mastery.

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