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Patterns of family management for adolescent and young adult brain tumor survivors.

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Patterns of Family Management for Adolescent and Young Adult Brain Tumor Survivors

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Abstract

Little is known about how families systemically incorporate the work of caring for adolescent and young adult (AYA) survivors of childhood brain tumors who often remain dependent on their families well into adulthood. The primary aim of this study was to develop a typology of family management (FM) patterns for AYA survivors. The secondary aims were to compare them to FM patterns previously described for children with chronic health conditions and to validate the patterns using quantitative and qualitative data. Guided by the Family Management Styles Framework, a sequential, mixed methods design was used to gather quantitative data from 186 mothers (primary caregivers) and 134 AYA survivors. FM patterns (Family-Focused; Somewhat Family-Focused; Somewhat Condition-Focused; and Condition-Focused) were identified using cluster analysis of data from the Family Management Measure. FM patterns were found to be similar to those for children with chronic health physical conditions and were significantly related to maternal quality of life, survivor quality of life (HRQL [self- and mother proxy report]), cancer-related variables (treatment intensity; medical late effects), and family functioning in theoretically meaningful ways. Significant demographic characteristics included private insurance and AYA survivors' engagement in school or employment. Qualitative analysis of data from 45 interviews with mothers from the larger sample provided additional support for and elaborated descriptions of FM patterns. Identification of FM patterns moves the science of family caregiving forward by aggregating data into a conceptually based typology thereby taking into account the complex intersection of the condition, the family, and condition management.

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These data have not been previously presented or shared.

Keywords

adolescents and young adults; oncology; family functioning; parenting; psychosocial functioning; mixed methods

Adolescent and young adult (AYA) survivors of childhood brain tumors are at increased risk for chronic health conditions and neurocognitive late effects (Armstrong et al., 2009; Birsko, Cohen, Dillon, Harvey, Krull, Klosky, 2016; Brinkman et al., 2016; King et al., 2016; Oeffinger et al., 2006). Therefore, survivors often experience decreased health-related quality of life (HRQL) and inability to achieve developmental milestones such as taking on the responsibilities for self-care, building social and romantic relationships, living independently, and being fully employed (Robison & Hudson, 2014). Primary caregivers, who are usually their mothers, are often challenged to assist and advocate for the functioning of their AYA survivors especially during the transition to adulthood (Forinder & Norberg, 2010; Hoven, Lannering, Gustafsson, & Boman, 2011; Palma et al., 2015; Woodgate, Taylor, Yanofsky, & Vanan, 2016).

Although some understanding exists regarding the role of maternal caregivers and psychosocial risk for survivors and their mothers (Berge & Patterson, 2004; Graf, Landolt, Mori, & Boltshauser, 2006; Rodenburg, Meijer, Dekovic, & Aldenkamp, 2006), less is known about family management (FM) or how families incorporate condition management into family life. To fill this gap, we conducted qualitative research (K. Knafl, Breitmayer, Gallo, & Zoeller, 1996) and developed the Family Management Style Framework (FMSF) (K. Knafl, Deatrick, & Havill, 2012). The conceptual components of the FMSF are contextual influences, definition of the situation, management behaviors, and perceived consequences. Based on the FMSF, we developed the Family Management Measure (FaMM) which is comprised of six scales or the key components of (FM) (K. Knafl, Deatrick, Gallo, Dixon, Grey, G. Knafl, & O'Malley, 2011).

Grounded in the assumption that families of children with chronic conditions develop distinct FM patterns (Rolland, 2005) and methods used to identify typologies in the family sciences (Mandara & Murray, 2002), a *k*-means cluster analysis of FaMM data was used to identify a typology of FM patterns for the aforementioned sample of children with chronic conditions and examine the relationship of these patterns to other variables (K. Knafl et al., 2013). We identified a typology of four FM patterns, Family-Focused; Somewhat Family-Focused; Somewhat Condition-Focused; and Condition-Focused, reflecting major differences in family response across the FaMM scales and the ease or difficulty families experienced in incorporating condition management related to their child's special needs into the family's everyday routine. Parents in the Family-Focused pattern minimized the difficulties associated with condition management and its impact on family life. Although acknowledging their child's special needs, they reported that they were confident in their ability to undertake the effort needed to manage the conditions while attending to family life. In contrast, parents in the Condition-Focused pattern indicated that condition management entailed considerable effort on their part and shaped everyday family life; they often questioned their ability to manage effectively. Condition-Focused parents shared a view of

family management (i.e. Mutuality) to a lesser degree than parents who were Family-Focused (K. Knafel et al., 2013). As hypothesized, AYA child adaptation and family functioning became progressively poorer (worsen) from Family-Focused to Condition-Focused (K. Knafel et al., 2013).

The primary aim of this study was to develop a typology of family management (FM) patterns for AYA survivors. The secondary aims were to compare them to FM patterns previously described for children with chronic health conditions and to validate the patterns using quantitative and qualitative data (Honea et al., 2008; Northouse, Katapodi, Song, Zhang, & Mood, 2010; Sherwood et al., 2012). We hypothesized that FM patterns for the AYA survivors of childhood brain tumors would be similar to those previously identified for children with chronic conditions. In addition, AYA HRQL and family functioning would become progressively poorer from Family-Focused to Condition-Focused. A sequential, mixed methods design (Creswell & Plano Clark, 2011) was used to identify, elaborate, and illustrate FM patterns. We used the same quantitative analytic strategies used in our previous study of children with chronic conditions (K. A. Knafel et al., 2013).

Method

Participants and Procedure

Data were gathered in a study of maternal caregivers, hereafter known as mothers, and their AYA childhood cancer survivors who lived with them (Barakat et al., 2015; Deatrick et al., 2014; Hobbie et al., 2016) and compared to quantitative patterns derived in the prior study of children with chronic conditions (K. Knafel et al., 2013). The study participants were eligible for the study if they spoke English and mothers self-identified and were identified by the survivor as primary caregivers. Survivors were (1) at least 5 years post diagnosis of a brain tumor in childhood and at least 2 years post treatment, (2) aged 14 through 40 years (Nass, Beaupin, & Demark-Wahnefried, 2015), (3) not married or living in a partnered relationship, (4) residing in the same household as their mother (5) judged by their mothers to be able to participate, and (6) not diagnosed with a genetic, brain-based condition or developmental delay prior to cancer.

Recruitment was conducted in an academic medical center in a large northeastern city. Institutional Review Board approved mailings were sent to 1,077 mothers identified in a large database of children with brain tumors treated in the past 30 years. In addition, 63 in-person contacts were made with mothers at neuro-oncology and survivorship outpatient clinics. For the quantitative phase, initial agreement for telephone screening was indicated by returning a reply card to the investigators provided in the mailing or in clinic, yielding willingness to be contacted by 384 mothers (35.7% response; 30% mailings; 90% clinic) and active refusals by 22 (2%). Of these, 190 (49.5%) mother-survivor pairs were eligible and 186 (98%) mothers and 134 (72%) survivors consented and provided complete data for the quantitative phase of the study

After screening, a telephone interview was scheduled with the mother and the survivor. Questionnaire data were collected by trained research assistants (RA) who read questions to the respondents and recorded responses into a data collection log. Data were subsequently

entered and reentered into a secure Access Data Base to check for accuracy. All agreed to be re-contacted for a follow-up qualitative home interview. One hundred eighteen (87%) lived within driving distance from the hospital. A diverse, purposive sample (Patton, 2002) was identified based on the Condition Management Effort scale of the FaMM and the Pediatric Oncology Quality of Life measure (mother proxy and survivor self-report) (Deatrick et al., 2014) by recoding maternal scores with their quartile membership. Mothers were selected ($n=56$) of which 52 mothers could be contacted; 3 mothers were ineligible; 1 mother declined; and 3 interviews could not be scheduled resulting in 45 mothers consenting and completing home interviews.

Measures (Sample items – See Supplementary Material)

Family Management—FM was measured by the Family Management Measure (FaMM), a 53-item instrument with 45 items for all parents and 8 additional items for partnered parents. The FaMM has 6 scales derived from a validation study with 579 parents of children with chronic conditions- excluding cancer and developmental delays. Internal consistency reliability in this study ranged from .72– .89. Higher scores on these scales mean a more normal family life and greater ease in condition management: (1) perceptions of the child's daily life; Child's Daily Life (CDL) (2) perceived competence to manage the condition; Condition Management Ability (Ability); and (3) perception of a partner having a shared view condition management (if applicable); Parental Mutuality (Mutuality). Higher scores on these scales mean a family life focused on condition management: (1) perception of demand associated with condition management; Condition Management Effort (Effort); (2) perception of problems associated with condition management affecting family life; Family Life Difficulty (Difficulty); and (3) perceptions of the condition impacting family life; View of Condition Impact (Impact) (K. Knafel, Deatrick, Gallo, Dixon, Grey, Knafel, & O'Malley, 2011).

Maternal Quality of Life—The Medical Outcomes Study: Short Form 36 Health Survey (SF-36) ($\alpha=.82$) is a normed measure of maternal health including physical functioning, bodily pain, role limitations due to physical health problems, role limitations due to personal or emotional problems, emotional well-being, social functioning, energy/fatigue, and general health perceptions (McHorney, Ware Jr., Rogers, Raczek, & Lu, 1992; Ware & Sherbourne, 1992). Items are scored so that a high score defines a more favorable health state on a 0 to 100 range.

AYA Quality of Life—The 15-item Pediatric Oncology Quality of Life (POQOL) measured both the mother's (proxy) perceptions about the survivor's physical HRQL ($\alpha=.89$) and emotional HRQL ($\alpha=.79$) and the survivor's perception about their physical HRQL ($\alpha=.74$) and emotional HRQL ($\alpha=.80$) (Bijtbeier et al., 2001; Goodwin, Boggs, & Graham-Pole, 1994; Kazak & Barakat, 1997). Higher scores indicate worse levels of HRQL.

Family Functioning—The 12-item General Functioning Scale of the Family Assessment Device (FAD) ($\alpha=.89$ for this sample) measured mother's perceptions of overall family functioning. Construct validity is well established with discrimination between nonclinical

and clinical samples (Epstein, Baldwin, Bishop, 1983; Kabacoff, Miller, Bishop, Epstein, & Keitner, 1990). Higher scores indicate worse levels of family functioning.

Cancer-Related Variables—The AYA survivor's treatment intensity (interrater reliability $\kappa=.97$) and medical late effects (interrater reliability $\kappa=.94$) were extracted from the medical record by a senior physician and nurse practitioner using the AYA Treatment Intensity Rating and Medical Sequelae adapted for brain tumor survivors (Kazak et al., 2012; Werba et al., 2007). A 5-point ordinal scale rates the treatment regimen from minimal (resection only) through most intensive (craniospinal radiation and intensive chemotherapy). A 4-point ordinal scale rates medical sequelae from minimum (no limitations) to severe restrictions (life threatening).

Qualitative Semi-Structured Interview—A semi-structured interview guide was constructed by three experts and modified after pilot testing with five families. The guide was organized using the FMSF to provide data that qualitatively describes and validates FM patterns.

Analysis Plan

Patterns of FM—Using the methods from our previous research (Dymanicki & Henry, 2012; K. Knafl et al., 2013; Henry, Tolan, & Gorman-Smith, 2005), *k*-means clustering of the FaMM scales and a *k*-fold likelihood cross-validation (LCV) (G. Knafl, Delucchi, Bova, Fennie, Ding, Williams, 2010) were conducted using SAS Version 9.4. The *k*-means clustering allows for missing values. Numbers of clusters were restricted to those with at least 10% of the observations in each to avoid sparse cluster solutions. Clusters were interpreted using their centroids (means of the FaMM scale values within each cluster). We then created a plot of normed cluster centroids for both AYA and the childhood chronic conditions, transforming them using the range 0–1 with larger normed values indicating more positive scale values.

Comparison of AYA FM Patterns with FM Patterns for Childhood Chronic Conditions—We compared FM patterns identified in the prior study of children with chronic conditions (K. Knafl et al., 2013) to those identified in this study. Differences in the number of families from each sample in each pattern were tested with the chi-square statistic. We then compared the plots of FaMM scale values for each sample to better understand them.

Validation of FM Patterns— χ^2 and one-way analysis of variance tests were used to test for differences in categorical and continuous demographics across FM patterns. Maternal demographics included: age, family income, education, employment status, insurance, race and ethnicity, and marital/partnered status. Survivor demographics included: age at data collection, gender, race and ethnicity, and time since diagnosis, and status (in school, working, or none). FM patterns were also validated by testing for significant differences in means scores of the six FaMM scales as well as of the mother proxy and survivor self-report of HRQL, professional ratings of cancer-related variables, and maternal reports of family

functioning. Comparisons were based on one-way analysis of variance (ANOVA) with post hoc analyses based on Duncan's multiple range tests.

Elaborate and Describe FM Patterns with Qualitative Data—To provide supplementary support for the FM patterns and provide illustrative case examples (Henry et al., 2005), two investigators coded the qualitative interviews using codes derived from the FMSF and FaMM scales and discussed any inconsistencies in their application of the codes to reach consensus. (For a detailed description of qualitative analyses see Palma et al., (2015)). A data summary matrix (Ayres, Kavanaugh, & K. Knafl, 2003) was then constructed that summarized data for each mother across all coding categories (K. Knafl, Deatrick, & Havill, 2012; K. Knafl et al., 2011). The data summaries for every mother within each FM pattern were then compared. Finally, narrative case summaries that are representations of two of the four patterns, Family-Focused and Condition-Focused, were developed.

Results

Demographic Characteristics

The sample is reported in Table 1 by inclusion in distinct study phases. While the total sample is 186 mothers and 134 survivors, 141 mothers were in the quantitative study only and 45 mothers were in both the quantitative and qualitative study phases. Demographic characteristics of mothers in the quantitative phase only and mothers in both the quantitative and qualitative phases, including age, education, employment status, insurance, race and ethnicity, and marital/partnered status, were not significantly different ($p > .05$). Survivors' demographics were representative of AYA brain tumor survivor samples in terms of age, gender, inability to work or go to school, and diagnoses (Armstrong et al., 2009).

Patterns of FM for AYA Childhood Brain Tumor Survivors

The intercorrelations of the FaMM scales (See Table 2) were almost all significant except View of Condition Impact and Parental Mutuality. They ranged in absolute value from .08 (weak)–.73 (moderate-between effort and difficulty). The k -fold LCV scores were best for four clusters using $k=5, 10, 15,$ and 20 folds, indicating that four clusters was a robust choice. For FM patterns based on the identified clusters, of the 186 families, 70 (38.0%) were Family-Focused, 62 (33.6%) were Somewhat Family-Focused, 31 (16.8%) were Condition-Focused, and 23 (12.5%) were Somewhat Condition-Focused. For the five FaMM scales reported by all mothers (Table 3), Family-Focused had the most positive averages (e.g., lowest difficulty average, highest ability average), Condition-Focused had the least positive averages (e.g., highest difficulty average, lowest ability average), and Somewhat Family-Focused and Somewhat Condition-Focused were in between with the second and third most positive averages respectively. Parental mutuality (which applies only for partnered mothers) was similar for Family-Focused and Somewhat Family-Focused as well as Condition-Focused and Somewhat Condition-Focused; Family-Focused and Somewhat Family-Focused had more positive values than Condition-Focused and Somewhat Condition-Focused. As shown in Figure 1, Family-Focused had especially high normed scores on Child's Daily Life, Condition Management Ability, Condition Management Effort,

Family Life Difficulty, and View of Condition Management Impact. Child Daily Life scores were relatively low for the three clusters. Parents in the Family-Focused and Somewhat Family Focused patterns had similarly high Mutuality scores in comparison to parents in the other patterns. Each of the four clusters generated distinctly different means for one or more of the FaMM scales supporting the need for all four clusters.

Comparison with FM Patterns for Childhood Chronic Conditions

FM patterns for AYA brain tumor survivors paralleled FM patterns derived in the prior study of children with chronic conditions (K. Knafel et al., 2013) but with different cluster centroids for the FaMM scales. As shown in Figure 1, the plots of both groups were similarly ordered in that Family-Focused had the most positive averages and Condition-Focused had the least positive averages. Somewhat Family-Focused and Somewhat Condition-Focused were in between with the second and third most positive averages respectively. The Somewhat Family-Focused, Somewhat Condition-Focused, and Condition-Focused families of children with chronic conditions reported better Child's Daily Life but more Difficulty than did the families of AYA brain tumor survivors with the same pattern. The normed scores for Impact were more similar across the same three AYA patterns than for those with childhood chronic conditions.

The percentage in each pattern differed significantly between the families of AYA brain tumor survivors and children with chronic conditions, $\chi^2(3) = 40.1, p < .001$. Families of AYA were more likely than families of children with chronic conditions to be Family-Focused (38% vs 24%) and Condition-Focused (17% vs 9%), less likely to be Somewhat Condition-Focused (12.5% vs 35%) and as likely to be Somewhat Family-Focused (32.6% vs 32.6%).

Validation of FM Patterns

Across demographics, FM patterns were significantly associated only with private insurance ($\chi^2(3) = 11.3, p = .010$) and AYA in school or employed ($\chi^2(3) = 8.23, p = .041$). Families with private insurance were more likely to be Family-Focused (47.5% versus 26.5%). Survivors who were in school or employed were more likely to be in families who were Family-Focused (48.7% versus 30.2%),

Validity of the FM patterns was supported by associations with measures of mother proxy report HRQL, survivor self-report HRQL, maternal health, and family functioning in expected directions ($p < .001$) (Table 4). Duncan's multiple range tests (post hoc analyses) identified that means for the validation measures were in the expected directions with means for all measures varying from more to less positive as patterns changed from Family-Focused to Somewhat Family-Focused to Somewhat Condition-Focused to Condition-Focused. As shown in Table 4, validity of the FM patterns was also supported by associations with treatment intensity ($p < .001$) and medical late effects ($p < .001$) ratings. In terms of treatment intensity, Family-Focused were more likely to have an AYA survivor with lower treatment intensity. Somewhat Family-Focused were just as likely to have treatment intensities similar to those for Family-Focused as to those for Somewhat Condition-Focused. Somewhat Condition-Focused and Condition-Focused were more likely to have an AYA who had more intense treatments. In terms of late effects, Family-Focused were less likely to

have an AYA survivor with severe medical late effects while Somewhat Family-Focused, Somewhat Conditioned-Focused, and Conditioned-Focused were more likely to have an AYA survivor with severe late effects.

Elaborate and Describe FM Patterns with Qualitative Data

Analysis of the qualitative data summary matrixes provided additional support for the FM patterns identified with the FaMM and validation measures. Mothers in the Family-focused pattern reported (1) having successful strategies for incorporating changes in survivor functioning into everyday family life; (2) family life was not negatively impacted despite profound stress related to AYA challenges; (3) AYA was perceived as essentially the same person he/she had been prior to having a brain tumor in ways that are important to the family; and (4) their families were able to manage, accommodate, and accept differences.

In contrast, reports by mothers in Condition-Focused families described (1) not having successful strategies for incorporating changes in survivor functioning into everyday family life; (2) family life being negatively impacted including experiencing profound stress related to AYA challenges; (3) AYA was perceived as a different person than he/she had been prior to having a brain tumor; and 4) their families being unable to manage, accommodate, or accept differences. Table 5 summarizes inductively derived thematic descriptions for the Family-Focused and Condition-Focused patterns and each FaMM factor. In addition, two cases (Amber whose family is Family-Focused and Nesta whose family is Condition-Focused) are provided to give examples of how each FM pattern and FaMM factor is reflected in both patterns.

Discussion

AYA childhood brain tumor survivors, their mothers who are often their primary caregivers, and their families are at risk for poorer adaptation than other childhood cancer survivors, their mothers, and families (Hoven et al, 2011). Little is understood, however, regarding the relationship of their adaptation and to how the family incorporates the management of their condition into family life (family management). To fill this gap in the literature, we have not only systematically built our model and methods over a 30-year period (K. Knafl & Deatrick, 1990; K. Knafl & Deatrick, 2003; K. Knafl et al., 2012) but we have systematically validated the results reported in this manuscript using quantitative plots, qualitative narratives (included in the original manuscript), and mixed methods strategies.

This study explored the patterns of FM for mothers of AYA childhood brain tumor survivors, the consistency of those patterns with those for children with other chronic conditions, and their validity using qualitative and quantitative evidence from multiple stakeholders (survivor, parent, professional). Four patterns of FM for AYA were identified that reflect distinct family responses with regard to the extent family life was focused on usual family routines and activities (Family-Focused Pattern) in contrast to those that focused on the demands of condition management (Condition-Focused pattern) (K. Knafl et al., 2013). In addition, the results confirmed that FM patterns present within families of AYA survivors of childhood brain tumors are similar to those identified for children with chronic health conditions. Centroids for each FaMM scale were ordered across the various FM patterns

both within the AYA typology and the chronic condition typology. Children with chronic conditions had better Child's Daily Life but more Difficulty in FM than AYA in Somewhat Family-Focused, Condition-Focused, and Condition Focused families. In addition, AYA had more similar Impact across the same three patterns than for childhood chronic conditions. Finally, AYA parents were more likely either to be Family-Focused or Condition-Focused than the childhood chronic conditions sample. Because of the chronic nature of the health conditions for both the children with chronic illnesses and the AYA survivors, the FM patterns were similar (Rolland, 2005). In addition, most likely because of different condition attributes (AYA neurocognitive late effects) for Somewhat Family-Focused, Somewhat Condition Focused, or Condition-Focused FM patterns, AYAs have less positive means or centroids for Child's Daily Life than children with chronic conditions and Impact was more similar for AYAs in the same patterns (Rolland, 2005). Finally, the qualitative data elaborated and illustrated the patterns and the typology thereby supporting their validity (Henry et al., 2005). In addition, the rich but specific descriptions provided a novel way to translate and disseminate the findings.

The results are consistent with other studies linking patterns to individual and family functioning (Dufour, Clément, Chamberland, & Dubeau, 2011; Farrelly, Cordova, Huang, Estrada, & Prado, 2013; Lindahl & Malik, 2011; Martinson et al., 2011; Ozono et al., 2010). While most prior research has focused on reports of other aspects of family life (e.g., family functioning, cohesion, conflict), this study focused on quantitatively derived FM patterns and studied their relationship to other aspects of mother and survivor functioning. Mothers in family-focused families reported significantly better maternal quality of life (maternal health); better family functioning; and, better survivor quality of life (HRQL) [self and mother proxy]. Professionals rated them as having less intense survivor cancer treatment and fewer cancer-related survivor late effects than Condition-Focused patterns. Furthermore, mothers from Family-Focused families reported better health, mothers and survivors reported better survivor HRQL, mothers reported better family functioning, and professionals rated less restricted survivor functioning.

In terms of demographics, significant findings for one family (private insurance) and one survivor characteristic (being either in school or employed) underscore the importance of current research related to these factors (Kuhlthau et al., 2016). When compared to other childhood cancer survivors, brain tumor survivors have the worst likelihood of being employed. While 18% of our total sample (Deatrick et al., 2014) was neither employed or in school, reports in the literature suggest that 25% of brain tumor survivors are unemployed (Kirchhoff et al., 2010; Kirchhoff et al., 2011), have low socioeconomic status (Ellenberg et al., 2009), and rely on governmental assistance. While the socioeconomic status of the family was not significantly different across the FM patterns, the survivor's own economic status was not queried.

This study was cross-sectional and yielded results that are most representative of white, married, non-Hispanic mothers who are educated, partnered, and have economic resources. The national incidence of brain tumors in Black children is lower than that of other racial groups, a phenomenon complicated by poorer survival rates, tumor histology, and age at diagnosis (Gurney, Smith, & Bunin, 1999). Even so, the self-selected sample for this study

did not contain a representative number of Black children and mothers (Central Brain Tumor Registry of the United States, 2010) and thus we were unable to examine the interplay of race and SES in FM. Future research can be also designed to better understand the contribution of other family members such as fathers or secondary caregivers. In addition, while a wide age range with inherent developmental differences is consistent with recommendations for AYA by the NCI regarding their definition of adolescence and young adulthood (NIH National Cancer Institute, April 23, 2017), it could have potentially attenuated the results of the study. The use of a single measure to create the typology, the FaMM, is challenged by Bogat and others (Bogat, Zarrett, Peck, & von Eye, 2012) who advocate for the use of additional variables in order to provide evidence that the patterns that comprise the typology are meaningfully different from each other. Finally, space restraints did not allow a more detailed presentation of qualitative results concerning the Somewhat Family-Focused and Somewhat Condition-Focused Patterns.

FM patterns reflect how families may organize to manage the condition, its comorbidities and complications, and emerging late effects, and therefore have relevance for enhancing the science of family caregiving by aggregating data from into a conceptually based typology. Because the FMSF integrates family processes, disease management, and outcomes, the resultant patterns take into account how complex characteristics of the condition and family intersect with condition management. The four FM patterns identify the challenges (FaMM scales) common across FM patterns and demonstrate the distinct ways in which challenges are managed across serious childhood health conditions and AYA brain tumor survivors. While other studies typically examine individual (e.g., HRQL) and family variables (e.g., family functioning), the contribution of this study is a closer examination of disease management and family life through a conceptual lens that typifies those key aspects of FM across health conditions and across multiple stakeholders (survivor, parent, and professional).

Since our long-term goal is to target or adapt an efficacious Family-Focused intervention, systematically organized qualitative data will be used to interpret the patterns for practice as well as to provide meaningful narratives for the intervention (Dymanicki & Henry, 2012)(p. 75). Typical educational interventions have concentrated on condition-related skills (i.e., “how to”) and not on strategies for integrating the skills into family life. Meaningful variables associated with FM patterns can be incorporated into future FM research; in particular, maternal health, survivor HRQL (proxy and self), survivor treatment intensity and late effects, survivor school or employment status, and family and survivor socio-economic status. By integrating condition-related care into ongoing family life, future interventions for Condition-Focused Families may hold unique promise.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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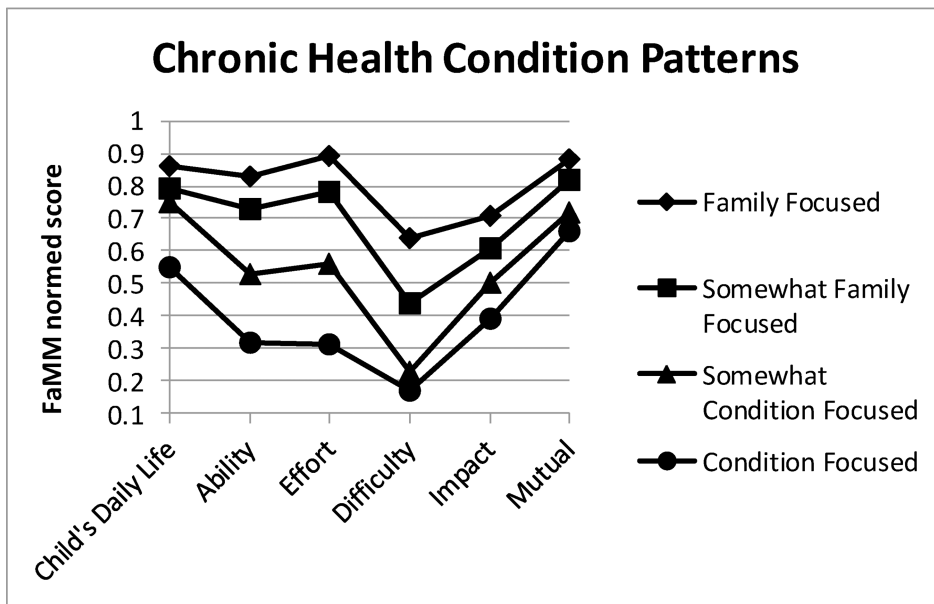
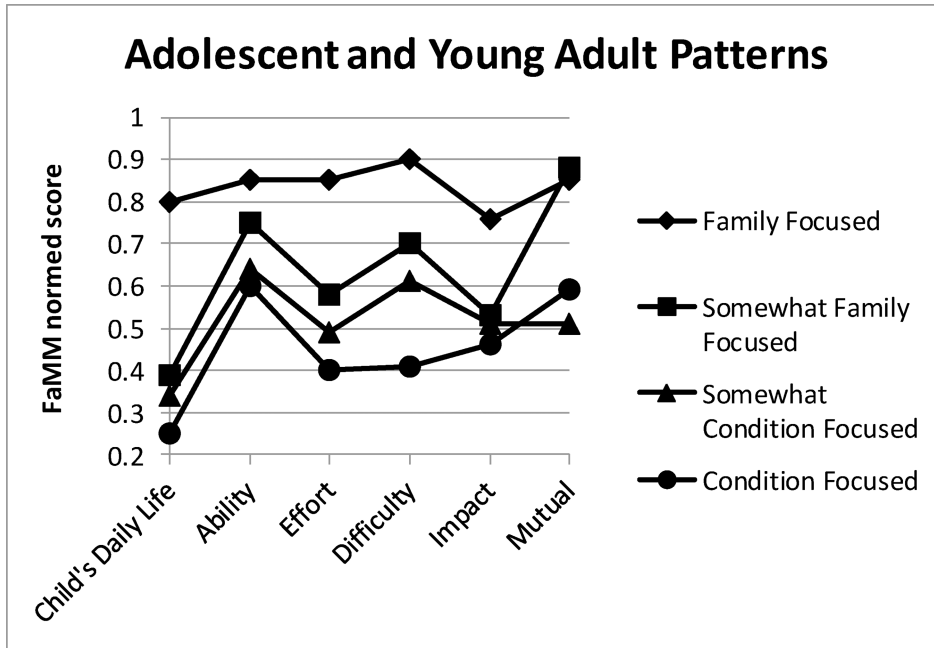


Figure 1. Normed scores for FaMM scales by family management patterns for adolescent and young adult families compared to chronic health condition families
Note: Each of the FaMM scales has been normed to be within the interval 0-1 but in such a way that a higher normed score means a more positive FaMM scale (larger for Child's Daily Life, Ability, and Mutuality and smaller for Effort, Difficulty, and Impact)

Table 1

Caregiver and Brain Tumor Survivor: Demographics and Cancer Related Characteristics in Quantitative and Qualitative Samples

Characteristic	Caregiver Quantitative Only N = 141	Caregiver Quantitative and Qualitative N = 45	Survivor Quantitative N = 134
Age in years			
Caregiver mean	51.89(<i>SD</i> = 6.02)	52.51(<i>SD</i> = 6.34)	52.1 (<i>SD</i> = 6.49)
Survivor mean	20.72 (<i>SD</i> = 4.49)	21.16 (<i>SD</i> = 4.83)	20.69 (<i>SD</i> =5.23)
Caregiver race/ethnicity			
White	127 (90.1%)	40(88.9%)	124 (92.5%)
Non-Hispanic	135 (95.7%)	44(97.8%)	131 (97.8%)
Caregiver highest education			
High school or less	34 (24.1%)	13 (28.9%)	28 (20.9%)
Some college	30 (21.3%)	10 (22.2%)	32 (23.9%)
College	39 (27.7%)	11 (24.4%)	35 (26.1%)
Some Grad School	10 (7.1%)	2 (4.4%)	10 (7.5%)
Graduate school	24 (17%)	7 (15.6%)	26 (19.4%)
Caregiver employment			
Full-time	71 (50.4%)	25 (55.6%)	58(68.7%)
Married or living with partner	76 (53.9%)	24 (53.3%)	76 (56.7%)
Years since diagnosis	12.97	13.15 (<i>SD</i> = 5.63)	13.08 (<i>SD</i> = 6.13)
Range	22	21	24
Survivor gender			
Male	78 (55.3%)	28 (62.2%)	76 (56.7%)
Female	63 (44.7%)	17 (37.6%)	58 (43.3%)
Survivor no school or work	27(19.1%)	8 (17.8%)	22(16.9%)
Survivor tumor location			
Posterior Fossa	56 (39.7%)	21 (46.7%)	50(37.3%)
Survivor tumor histology			
Low grade glioma	69(49%)	19(42%)	73(54%)
Medulloblastoma	42(30%)	16(36%)	32(24%)
Moderate restrictions to daily living	74(55.5%)	25 (55.6%)	59 (44%)
Treatment intensity			
1. Resection only	41 (29.1%)	12 (26.1%)	62(33.3%)
2. Focal radiation ± non-intensive chemotherapy	42(29.8%)	14 (31.1%)	55(29.6%)
3. Moderate chemotherapy ± focal radiation, but no craniospinal radiation	14(9.9%)	2 (4.4%)	19(10.2%)
4. Craniospinal radiation ± non-intensive chemotherapy or HDC with stem cell rescue	40(28.4%)	15 (33.3%)	45(24.2%)

Note: Sample characteristics for children with chronic conditions described elsewhere (Knafl, et al., 2011; Knafl, et al., 2013).

Table 2

Pairwise Correlations between Family Management Measure Scales

	Child's Daily Life	Ability	Effort	Difficulty	Impact
Ability	0.54**				
Effort	-0.64**	-0.50**			
Difficulty	-0.70**	-0.68**	0.73**		
Impact	-0.63**	-0.59**	0.62**	0.67**	
Mutual	0.18*	0.37**	-0.18*	-0.43**	-0.08

* $p < .05$

** $p < .001$

Table 3
Quantitative Results: Four Family Management Patterns for Brain Tumor Survivors – Mean Scale Scores and Standard Deviation

Scale ^a (Possible Range)	Family Management Pattern Mean Values ^b					<i>F</i> ^c
	Family-focused	Somewhat Family-Focused	Somewhat Condition-Focused	Condition-Focused	Standard Deviation	
Child's Daily Life (5–25)	20.9 _A	12.7 _B	11.7 _B	9.9 _C	3.8	<i>F</i> (3,179)=86.4
Ability (12–60)	52.8 _A	47.8 _B	42.5 _C	40.8 _C	5.1	<i>F</i> (3,179)=49.9
Effort (4–20)	6.4 _A	10.8 _B	12.2 _C	13.6 _B	3.0	<i>F</i> (3,180)=53.7
Difficulty (14–70)	19.4 _A	30.7 _B	35.9 _C	47.3 _D	4.5	<i>F</i> (3,179)=290.3
Impact (10–50)	19.7 _A	29.0 _{A,B}	29.7 _B	31.7 _C	5.1	<i>F</i> (3,179)=58.8
Mutual (8–40)	35.1 _A	36.3 _A	24.3 _B	26.9 _B	5.4	<i>F</i> (3,141)=32.9

^aFor three scales: Child's Daily Life (CDL), Condition Management Ability (Ability), Parental Mutuality (Mutuality) higher scores are indicative of a more normal family life and greater ease in condition management; for three other scales: Condition Management Effort (Effort), Family Life Difficulty (Difficulty), View of Condition Impact (Impact), higher scores are indicative of a family life focused on the difficult work of managing the condition and associated issues.

^bMeans with different upper case subscripts are jointly significantly ($p < .05$) different while means with the same subscripts are not using Duncan's multiple range test.

^cAll *F* tests significant at $p < .001$.

Table 4
 Family Management Patterns: Validation with Mean Family, Caregiver and Child Functioning as Well as Treatment Intensity and Medical Late Effects Ratings across Four Patterns of Family Management

Functioning ^b	Family Management Pattern Mean Values ^a				F ^c
	Family-Focused	Somewhat Family-Focused	Somewhat Condition-Focused	Condition-Focused	
Family Functioning (range 1–4 [better to worse])	1.6 _A	1.7 _A	2.0 _B	2.0 _B	<i>F</i> (3,180)=12.1
Caregiver Health (range 0–100 [better to worse])	79 _A	72.6 _{A,B}	65.4 _B	63.8 _B	<i>F</i> (3,180)=5.8
Survivor HRQOL Physical Proxy (range 9–63 [better to worse])	14.9 _A	28.8 _B	30.4 _B	36.5 _C	<i>F</i> (3,180)=45.8
Survivor HRQOL Physical (range 9–63 [better to worse])	10.3 _A	12.4 _{A,B}	14.1 _{B,C}	17.2 _C	<i>F</i> (3,121)=6.9
Survivor HRQOL Emotional Proxy (range 6–42 [better to worse])	9.7 _A	14.4 _B	18.1 _C	20.7 _C	<i>F</i> (3,180)=22.1
Survivor HRQOL Emotional (range 6–42 [better to worse])	17.4 _A	21.3 _A	27.6 _B	27.9 _B	<i>F</i> (3,121)=9.2
Treatment Intensity Rating (1–5 [better to worse])	1.9 _A	2.4 _{A,B}	2.6 _B	2.8 _B	<i>F</i> (3,180)=9.2
Medical Late Effects Rating (1–4 [better to worse])	1.9 _A	2.8 _B	2.9 _B	2.9 _B	<i>F</i> (3,180)=23.9

^a Means with different upper case subscripts are jointly significantly ($p < .05$) different while means with the same subscripts are not using Duncan's multiple range test.

^b Family Functioning = the General Functioning Scale of the Family Assessment Device; Caregiver Health = the Medical Outcomes Study Short Form 36 Health Scale; HRQOL = Pediatric Oncology Quality of Life

^c Standard analysis of variance models with constant standard deviations.

^c All *F* tests significant at $p < .001$.

Table 5

Thematic Descriptions and Narrative Case Descriptions of Family-Focused and Condition-Focused Family Management

Family Management	Thematic Description –Family Focused Case	Narrative Case Description - Family-Focused Case-Amber	Thematic Description –Condition-Focused Case	Narrative Case Description - Condition-Focused Case=Nesta
Overall Description	Overall, families were characterized as positive about meeting past, present and potential future challenges related to condition management and child's special needs.	Amber is a 25-year-old female survivor of Medulloblastoma diagnosed at age 9 and treated with surgery and intensive craniospinal radiation. Amber has moderate restrictions of daily activity, visible scarring/asymmetry, significant learning/intellectual disabilities with special education placement, Type 2 Diabetes, and uses growth hormone. She lives at home with her mother and father; her siblings are now living outside the home. She graduated from college but employment opportunities are limited to substitute teaching. Parents reluctantly applied for social security disability benefits for her.	Overall, families were characterized as having considerable difficulty meeting the challenges related to condition management and their children's special needs.	Nesta is a 14-year-old male survivor of a craniopharyngioma, diagnosed at age 5 and treated with surgery only. He has moderate restrictions to his daily activities, mild visual impairment, mild to moderate hearing impairment, thyroid disorder, learning disorder, psychological problems, panhypopituitarism and pseudoseizures. Mother described him as requiring constant monitoring, isolated with no friends, and immature for his age. He is aided by a part-time resource room and tutor in school. His older siblings live in the home with the survivor, mother and stepfather.
Child's Daily Life-FaMM Factor	Acknowledged both child's strengths and vulnerabilities and provided examples of both. Strengths were emphasized over vulnerabilities. Children are described as having meaningful activities, despite the need for some special supports.	Amber's mother stated, "...her IQ test came out average, which to me was fine after what the kid's been through. And she had a lot of strengths...Some of the weaknesses were nothing new to me. Processing slower...." Also, her mother noted that although Amber has a college degree, she had significant assistance from college and family members.	Children described in terms of vulnerabilities, with emphasis on differences from peers. Children are viewed as having few meaningful activities and as unhappy.	Nesta's mother focused on vulnerabilities and differences from peers. For example, she said "...food is 24/7...he's never been happy because it would mean he'd need to stop thinking about food... he has an outburst...it's over and he doesn't think about it" and "I accept he's different but I'm not willing to accept, that there's not more for him."
Condition Management Ability-FaMM Factor	Acknowledged caregiving can be difficult, but noted ways it had become less difficult over time. Emphasized competence and pride in ability to manage condition and	Amber's mother said that she felt competent to meet caregiving demands related to Amber's special needs pointing to routines to help her daughter care for herself, "...at	Recognized some areas of competence, but described ongoing misgivings about management ability. Described how management strategies have become less successful over time.	While Nesta's mother reported that, "...the temper tantrums over food have gotten better as he's gotten older" overall she did not see his condition as manageable in terms of her competency to carry

Family Management	Thematic Description –Family Focused Case	Narrative Case Description - Family-Focused Case-Amber	Thematic Description –Condition-Focused Case	Narrative Case Description - Condition-Focused Case=Nesta
	children’s special needs. Described goals for adapting management approach their children matures.	night I push things she needs (like medications) to the front of the counter because I don’t want her to forget. That way I can monitor how she is doing and not too intrusive.”		out management now and into the future. “It’s just frustrating, because there has to be an answer. I can’t find it.”
Condition Management Effort-FaMM Factor	Described selected areas of effort that remain problematic, but also strategies for transferring management responsibility to their children. Acknowledged her own reluctance to transfer full responsibility to their children.	Amber’s mother recognized the importance of the survivor taking more responsibility for condition management. However, she also reported that it was sometimes difficult to refrain from doing things for her, such as refilling prescriptions instead of encouraging Amber to call for renewals herself, “I know it is more work but I have to keep the long term objective in mind.”	Described considerable effort that needs to be directed to caring for their children and ongoing challenges. Caregiving is described as unrelenting and undertaken in the context of limited resources and supports.	Nesta’s mother described not only his 24-hour, 7 days/week care that included locking anything in the kitchen with food in it, but also the work of asking professionals questions, which they could not answer. She described constantly struggling with knowing how much to limit her child’s eating, “he needs 24/7 care, otherwise he will leave to get food.”
Family Life Difficulty-FaMM Factor	Described many positive aspects of family life, contrasting it to difficulties in the past. Described successful efforts to maintain a normal family life and support from extended family.	Family life was viewed positively, but difficulties were acknowledged. Amber’s mother stated, “So there was changes both ways, some we became better parents because of what we went through and then other things we had to work around.”	Described family life as unsatisfying and focused on condition management and the difficult challenges associated with their children’s special needs. Described anticipating more problems in the future.	Mother reported that family life changed dramatically after Nesta’s diagnosis, with many aspects of everyday life becoming more difficult. The condition and its management had become the center of family life, especially for siblings. Siblings were described as both protective and resentful, “...it is very hard when you can’t get an adult to understand, [how can you] expect children (siblings) to understand?”
View of Condition Impact-FaMM Factor	Described ways the condition had changed family life, but also how they have maintained a hopeful outlook for their children’s and families’ futures. Described lingering concerns about possible late effects.	Amber’s mother acknowledged the seriousness of the situation and implications for her daughter and family’s future. Although the health care team advised that it is unlikely Amber will live independently, she continued to weigh evidence supporting and contradicting that probability. Meanwhile she focused on Amber being more independent, “I just	Described both the current impact of the condition on their children and families and the belief that the negative consequences of the brain tumor will continue and likely increase in the future.	For example, Nesta’s mother addressed the seriousness of his condition and its profound implications for her son’s and family’s future, “The older he gets, we realize I am a full-time caregiver probably for the rest of his life...where that does that leave us?”

Family Management	Thematic Description –Family Focused Case	Narrative Case Description - Family-Focused Case-Amber	Thematic Description –Condition-Focused Case	Narrative Case Description - Condition-Focused Case=Nesta
Parental Mutuality-FaMM Factor	Emphasized how they and their husbands/ partners have worked together and supported each other.	want her to do something where she feels valuable and in control and I'm not telling her she wants to go out and go find an apartment now but I'm sure at some point she's gonna want to do that..." She described her relationship with her husband as complementary in meeting challenges they faced, "If I was feelin' like this, he happened to be feeling like [that]... we never pulled each other down..."	Described their husbands/ partners as not fully recognizing the many challenges they have faced over the years.	Nesta's step-father was sympathetic to the many caregiving challenges; however, his mother said that he had not fully understood them until spending more time at home due to a recent illness. "I said, well, that's what I would tell you when you worked late."

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