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How parents cope with their child's diagnosis and treatment of an embryonal tumor: Results of a prospective and longitudinal study

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Abstract

Purpose—The current study reports longitudinal coping responses among parents of children diagnosed with an embryonal brain tumor.

Patients and Methods—Patients (n=219) were enrolled on a treatment protocol for a pediatric embryonal brain tumor. Their parents (n=251) completed the Coping Response Inventory at time of their child's diagnosis and yearly thereafter, resulting in 502 observations. Outcomes were examined with patient and parent age at diagnosis, patient risk, parent gender and education as covariates.

Results—At the time of diagnosis, the highest observed coping method was seeking guidance with well above average scores (T=61.6). Over time, younger parents were found to seek guidance at a significantly higher rate than older parents (p=.016) and the use of acceptance resignation and seeking alternative results by all parents significantly increased (p=.011 and p<.0001 respectively). The use of emotional discharge was also observed above average at time of diagnosis (T= 55.4) with younger fathers being more likely to exhibit emotional discharge than older fathers (p=.002). Differences in coping according to age of the patient and parent education level are also discussed.

Discussion—Results show a high need for guidance, and above average emotional discharge, especially among younger parents. It is imperative for the healthcare team to lead with accurate information so that these parents may make informed decisions about the care of their child. This need remains high years after diagnosis. Therefore it is critical to continue a consistent level of effective communication and support, even following treatment.

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Keywords

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The diagnosis of a pediatric embryonal brain tumor is one of the most disruptive and stress producing events that a parent could experience. It also holds the potential for long lasting psychological impact.¹ Diagnosis launches the family into an overwhelming number of critical decisions, each with varied consequences and each producing a unique source of potential stress. Lengthy treatment periods are followed by years of necessary follow-up making pediatric cancer more like an ever-changing unremitting disease rather than an acute medical condition. However, not all parents are at risk for developing psychological sequelae. Coping methods utilized by the parents may determine the observed differential response.²

The aggressive nature of an embryonal brain tumor requires several stages of treatment including surgical resection, risk-adapted craniospinal irradiation, and adjuvant chemotherapy.³ Each stage is accompanied by potential short and long-term side-effects that must be considered when making treatment decisions. Over time treatment presents new and continuing caregiver demands on the family. The phase of active treatment can be as long as one year and many families are required to relocate or separate in order to complete treatment. The requirement to be apart is a significant source of stress independent from treatment.⁴ Many parents report a reduction in quality of life.⁵ Lengthy absences from employment can also produce financial strain with far reaching implications.⁴ When treatment is complete many families begin to refocus on family life and patients reintegrate into the school system and social activities. Unfortunately this is also when concerns about their child's future well-being and treatment related sequelae begin to surface.⁶⁻⁸ The transition away from active treatment is when many families face unrelenting fears of relapse bringing a level of uncertainty that can prove very difficult to resolve.^{1,4,6,9,10}

Researchers have two orientations of coping: 1) problem-focused and 2) emotion-focused).^{11,12} Problem-focused orientation is considered to be action oriented by dealing directly with the problem at hand, and attempting to act in order to make change.¹¹ This is referred to as approach coping. In contrast, emotion-focused coping tends to be more "avoidant" by mitigating the stress even though the actual conditions have not changed^{11,12} and has been associated with increased risk for depressive and anxiety symptoms.²

Studies that examine the coping responses are rare and tend to depend on qualitative data, frequently employing an interview format.¹³ There is a need for future research to describe how parents cope according to the phase of treatment and time of recovery.¹ The current study reports on the prospective examination of coping responses among parents of children treated for a pediatric embryonal brain tumor from the time of diagnosis onward, for a 5 year period. This methodology allows identification of coping patterns over time, potential maladaptive strategies and possible areas to intervene, important to the long term psychological health of the family.

Materials and Methods

Patient Population

Study participants were recruited from a multi-site IRB-approved clinical trial for patients who are 3 years of age or older and newly diagnosed with an embryonal brain tumor. Written consent was obtained for participation. At time of the study, patients (n=290) diagnosed with medulloblastoma, primitive neuroectodermal tumor, pineoblastoma or

atypical rhabdoid tumor were enrolled and treated at 1 of 9 separate institutions: St. Jude Children's Research Hospital (Memphis, TN; n=144), Duke University Medical Center (Durham, NC; n=20), Royal Children's Hospital of Brisbane (Australia; n=32), Texas Children's Hospital (Houston, TX; n=31), The Hospital for Sick Children (Toronto, Canada; n=24), Royal Children's Hospital of Melbourne (Australia; n=12), Sydney Children's Hospital (Australia; n=9), and Children's Hospital of Philadelphia (Philadelphia, PA; n=7), and Children's Hospital of Westmead (Sydney, Australia; n=11). The last site did not participate in collecting parent coping data for the current study. Patients were included in the current study if at least one of their parents had completed a questionnaire measuring coping in response to their child's diagnosis and subsequent treatment (N=219).

Patients were treated with post-surgical risk-adapted craniospinal irradiation (CSI) followed by 4 cycles of adjuvant chemotherapy (cyclophosphamide, cisplatin, vincristine) with stem cell support (males=131, females=88). Average-risk (AR, n=152) patients received 23.4 Gy CSI, 3-D conformal boost to the primary site to a total dose of 55.8 Gy. High risk (HR, n=67) patients received 36–39.6 Gy CSI and 3D conformal boost to the primary site to a total of 55.8–59.4 Gy. The median age at diagnosis of the patient group was 8.55 years (M=9.14 years, SD=4.01 years) and they were an average of 2.61 years from diagnosis (SD = 1.57) at the time of their most recent follow-up. The majority of the patients were White (78.5%), with 8.6% Black, and 4.1% Asian. Remaining patients were either of Unknown race or classified as Other.

Parent Population & Assessment of Coping

Parents were asked to complete the 48-item Coping Response Inventory- Adult Form (CRI-A)¹⁴ at time of their child's diagnosis and yearly thereafter, providing longitudinal measures of 8 coping responses (Table 1). The CRI-A was developed with normative sampling to measure a person's coping response to a specific stressful life circumstance, and monitor stability and/or change in coping responses over time. To facilitate comparison among scales raw scores each of the 8 subtests were converted to standardized T-scores (M = 50, SD = 10). Coping responses resulting in T-scores ranging from 46–54 are considered within "Average" frequency, while those from 55–59 are "Somewhat above Average". T-scores 60–65 are considered "Well Above Average" and those >66 are "Considerably Above Average".¹²

Two hundred mothers and 51 fathers participated, resulting in 502 observations over a 6 year period. The majority of the parents were White (76.9%), with 8.1% reported as being Black. Remaining parents were classified as Other. At the time of diagnosis 78.8% of the parents were Married, 10% were divorced, 6% separated, and 5.2% declared they were single. Also at the time of their child's diagnosis, parents ranged from 19 to 58 years of age (Median=37 years) and had 8 to 23 years of education (Median=14 years).

Statistical Analysis

Random coefficient models were used to estimate the longitudinal change in each of the eight coping responses while taking into account the correlations of the within-subject repeated observations.¹⁵ Random coefficient models are regression models for repeated measures that allow the intercept and slope for each subject to be a random deviation from the population intercept and slope. The effect of sociodemographic and clinical factors on the estimated intercept and slope for each coping response were investigated using similar models. Factors included patient age at diagnosis (years), patient risk (average or high), parent age at diagnosis (years), parent gender and number of years of education completed by the parent. For each coping response, single factor models were built to include the factor main effect, time on study, and the interaction of the factor with time on study. Factors with

either a significant main effect or a significant interaction at the 0.10 alpha level in the single factor models were further investigated using multivariable models to explore which factors were significantly correlated with the estimated intercept and/or slope after adjusting for other factors. The Akaike Information Criterion (AIC) was used to find the best fitting and most parsimonious model. All models were analyzed using PROC MIXED in SAS Release 9.2(SAS Institute, Cary, NC). All reported *P*-values are two-sided and are not adjusted for multiple comparisons.

Results

At time of diagnosis parents were observed to be more likely to use approach coping methods, rather than avoidant, in response to their child's diagnosis and treatment (Figure 1). The highest endorsed coping method was seeking guidance which was well above average (Mean $T=61.6$, $SD=7.7$). Positive reappraisal and emotional discharge were somewhat above average (Mean $T=55.0$, $SD=8.2$ and Mean $T=55.4$, $SD=8.8$ respectively) and problem solving was on the border (Mean $T=54.9$, $SD=8.3$). The mean values for the other coping mechanisms were in the average range at the time of the child's diagnosis (Figure 1).

Longitudinal models showed that coping by seeking guidance, positive reappraisal, problem solving and emotional discharge remained above average over the entire study period with no significant change over time. There was a significant increase in parent acceptance and resignation ($p=.011$) as time on study increased. Coping by seeking alternative results appeared to increase in the first year after diagnosis and then level off. However, linear analysis showed a significant increase over time from diagnosis ($p<.0001$). Logical analysis and cognitive avoidance was average or below average at the time of diagnosis and showed no significant change over time (Figure 2).

Multivariate modeling found that parents with higher levels of education have significantly higher logical analysis scores at the time of their child's diagnosis ($p=.004$), and their use of this coping method decreases at a significantly slower rate than those with lower levels of education over the study period ($p=.043$). Parents with a higher level of education also have significantly higher seeking guidance scores than parents with lower years of education at the time of their child's diagnosis ($p=.034$) and these differences do not significantly change over time.

Parents with children who were older at the time of their diagnosis scored significantly higher on positive reappraisal than parents of younger children ($p=.019$), and these differences seen at diagnosis remained over time. Younger parents are more likely to use cognitive avoidance in response to their child's illness, than are older parents ($p=.003$). Over time, younger parents also seek guidance at a significantly higher rate than those who are older ($p=.017$).

Both mothers and fathers show using problem solving as a method of coping that is somewhat above average at the time of diagnosis. However, while the fathers' rate of problem solving did not significantly change over time, mothers show a significant increase in the use of problem solving methods over time ($p=.015$). While there was little difference between the use of emotional discharge at the time of diagnosis among mothers based on age (both older and younger were somewhat above average), the older the father was the less likely it was that he would use emotional discharge as a way to respond to his child's illness ($p=.019$). Young fathers were as likely to use emotional discharge as the young mothers.

Given this was a multi-site study, location of enrollment was also considered as a possible covariate. No significant differences were found with regard to outcome by study site.

Coping responses among parents whose child had experienced posterior fossa syndrome (n=37) were compared to parents whose child did not. No significant differences in coping between these two groups were found.

Discussion

The current study is the first to quantitatively and longitudinally examine coping responses of parents who had children diagnosed and treated for a brain tumor. At the time of diagnosis all parents showed a very high need for seeking guidance, and an above average use of emotional discharge. These coping responses remained at above average levels over the course of the study period, including periods of active treatment, transition, and post-therapy follow-up. These tendencies were also especially true for younger parents.

The results emphasize a clear need by these parents for detailed information and access to resources that will help them in their quest for informed guidance. At the time of diagnosis it is understandable that parents of children diagnosed with a brain tumor could be quickly overwhelmed while making the many critical decisions necessary for their child's care. It is imperative for the health care team to provide open, honest, and accurate communication so that the parents may fulfill their need to cope by being informed and active in their child's medical care. Parents were found to be satisfied when communication was open and honest, and when health care providers gave them time to express their feelings and answer their questions.¹⁶ These needs remained high over time, especially for younger parents. Therefore it is also important to continue a consistent level of effective communication, even subsequent to active treatment.

Previous research has shown that parents access a variety of sources including mass media and the internet.¹⁷ Online synopses of research findings and/or links to up to original information on treatment related issues, post-treatment transition, and treatment related sequelae could provide the information these families seek. If posted by a reliable source, such as organizations formed to support those affected by a brain tumor, parents could be certain that they are receiving the credible information required to make informed choices. This in turn could help raise their confidence in making decisions for the care of their child.

In addition to seeking guidance from professionals, friends, and family, the seeking guidance subtest also includes endorsement of an item with regard to praying for direction. Turning to one's faith for guidance has been described as an important source of comfort for parents.¹⁸ It also confirms a role for spirituality and religion in parental coping and represents prayer as an active coping response, rather than a passive plea. This result poses questions not able to be addressed in the current study and therefore also represents an inherent limitation. However, in the current study the parents' belief in the power of this prayer and the efficacy of such action is unclear. Prayer is a highly personal and variable behavior. Therefore, it is important for the health care team to understand the parents need or lack thereof, for prayer as a way to cope, make available the opportunity to pray if desired, and offer contact with hospital chaplain as appropriate.

Parents used problem solving as a way to cope at a somewhat above average rate at time of diagnosis, and this response remained above average over time. While fathers' use of problem solving remained consistent over time, mothers were found to have a significant increase in problem solving behavior over the study period. This result may reflect a difference in parenting roles while caring for the child during treatment, transition and periods of post-therapy follow-up. It is possible that more mothers than fathers take on the

primary caregiver role and therefore find themselves in a position of advocating for their child and making an increased number of decisions with regard to their child's illness, making more problem solving responses than the fathers. Effective problem solving skills are an important consideration for positive coping among primary caregivers. A multi-site study involving children with mixed cancers reported that problem solving skills training intervention for mothers of children with cancer, including eight one-hour individual and standardized sessions, resulted in reduced negative affect, avoidance style, and negative problem orientation when compared to those mothers who did not receive the intervention.¹⁹ This study also raised questions with regard to the optimal timing of such an intervention. The authors found that mothers refused participation in the intervention at twice the rate within the first 4 weeks after diagnosis than after 4 weeks. While several potential moderating variables were examined, child diagnosis was not. Future research to evaluate the refusal rate and efficacy of the intervention by child diagnosis would indicate the potential of problem-solving skills training to help support positive coping for parents of children with variable disease, treatment plans, and recovery patterns.

At the time of diagnosis parents reported an above average use of emotional discharge as a way to cope with their child's diagnosis and treatment. The emotional discharge remained above average over the study period and included behaviors such as shouting to let off steam, crying, taking anger or depression out on others, or avoiding others. These behaviors are considered avoidant coping responses and emotion-focused. They reflect attempts to avoid thinking about or actively solving the problem. While crying may provide an emotional catharsis, the problem has not been altered by doing so. While these responses by the parent may be understandable given their child has been diagnosed and treated for a life threatening disease, the fact that emotional discharge remains above average over time is cause for concern. The emotional health of the parent and other family members may deteriorate to include higher levels of depression or anxiety.^{1,13,20} Family based intervention may be necessary in order to improve adjustment for those who continue to utilize emotional discharge as a way to cope.

Parents exhibited average use of 'seeking alternative results' and 'acceptance and resignation' at time of diagnosis. Results indicated a significant increase in these two coping responses over time from diagnosis. Items included in the seeking alternative results subtest include trying to make new friends, trying to help others deal with a similar problem, reading more for enjoyment, and turning to work or other activities to help manage the stressor. Items included acceptance and resignation include losing hope that things would ever be the same, feeling like time would make a difference, realizing you have no control over the situation, accepting the problem, and expecting the worst possible outcome.

While both seeking alternative results and acceptance and resignation are considered avoidant, the adaptive value of coping responses should be considered according to the phase of cancer¹ because these responses may be realistic for parents given their particular situation and environment. Many times these parents have been taken away from their regular support system and find themselves needing to make new friends among other parents facing similar challenges. Time waiting at the hospital and appointments lends itself to reading or other activities that can help relieve some of the cognitive and emotional burden, even for just a short time. Time often does make a difference in the outcome of their child's disease and accepting the situation may be important for the parent to move forward through transition and post-therapy follow-up. An honest assessment would logically lead to the conclusion that things will not ever be the same for their child, themselves, or their family. The diagnosis of a pediatric brain tumor forever changes the course of a family and resets their definition of "normal." Therefore the responses described could be viewed as necessary for effective coping and a healthy adjustment to the circumstances.

In summary, the present results emphasize that understanding how parents cope with their child's diagnosis, and supporting their needs for effective coping, are important responsibilities of the health care team. Support organizations, chaplain services, and members of ancillary care teams can also play important roles. Opportunities for accurate and informed guidance, emotional release, and effective problem solving skills training are critical to parents' adjustment and health.

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Figure 1. Observed coping responses by all parents at time of their child's diagnosis is (T-scores $M=50$, $SD=10$).

Figure 2. Predicted parent coping responses by time since diagnosis (T-scores $M=50$, $SD=10$).

Table 1

Coping Response Inventory scales and descriptions. (from Moos, R.H. 1993)

Approach Coping Responses

Logical Analysis	Cognitive attempts to understand and prepare in response to their child's diagnosis and treatment and related consequences
Positive Reappraisal	Cognitive attempts to restructure their child's diagnosis and treatment in a positive way while still accepting the reality of the situation
Seeking Guidance and Support	Behavioral attempts to seek information, guidance or support
Problem Solving	Behavioral attempts to take action to deal directly with their child's diagnosis and treatment

Avoidant Coping Responses

Cognitive Avoidance	Cognitive attempts to avoid thinking realistically about their child's diagnosis and treatment
Acceptance or Resignation	Cognitive attempts to react to their child's diagnosis and treatment by accepting and resigning to the illness
Seeking Alternative Rewards	Behavioral attempts to get involved in substitute activities to create new sources of satisfaction
Emotional Discharge	Behavioral attempts to reduce tension by expressing negative feelings