De-escalation of Therapy for Pediatric Medulloblastoma: Trade-Offs Between Quality of Life and Survival

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RESULTS. Parents and providers showed differences in which factors they believe have the greatest impact on quality of life for children with medulloblastoma and their families. For parents, the most important factor is social functioning and their child’s ability to make friends and have a social life. In contrast, providers thought that parents cared most about their child’s cognitive functioning and ability to attend and perform in school. Conclusion. Understanding parents’ perspectives on quality of life is important in terms of providing support services that target the areas that the parents prioritize. The types of functioning that are most strongly correlated with quality of life from the parents’ perspective may be the ones that should be targeted to protect during treatment. Pediatr Blood Cancer 2014;61:1300–1304. © 2014 Wiley Periodicals, Inc.

**Background.** Treatment intensity for pediatric medulloblastoma may vary depending on the type of medulloblastoma. In some cases, the dose of radiation may be reduced or eliminated. Correspondingly, there may be trade-offs between quality of life and survival. In this study, focus groups were conducted with parents and clinicians to explore their opinions about these trade-offs as well as the alignment/misalignment between parents and clinicians regarding the trade-offs. Methods. One hour semi-structured focus groups were conducted with parents of children with medulloblastoma and health care providers who were involved in the care of these children. Results. Parents and providers showed differences in which factors they believe have the greatest impact on quality of life for children with medulloblastoma and their families. For parents, the most important factor is social functioning and their child’s ability to make friends and have a social life. In contrast, providers thought that parents cared most about their child’s cognitive functioning and ability to attend and perform in school.

**METHODS**

Semi-structured focus groups were conducted with parents of children with medulloblastoma and health care providers who were involved in the care of these children. Patients were identified through the neuro-oncology database and their parents (father and/or mother) were invited to participate in the focus groups via an invitation letter (196 parents and 32 providers were approached to take part of our study.)

Parents who expressed interest in the focus group were contacted directly by research personnel. Groups were stratified with parents and providers participating in separate sessions. These 1 hour focus groups were conducted at The Hospital for Sick Children (SickKids) in Toronto, and at British Columbia Children’s Hospital (BCCH) in Vancouver, Canada. Discussions were led by a medical anthropologist with 10 years of experience running focus groups. All discussions were audio recorded and transcribed. Participants were asked their opinions on the factors that most affect quality of life. They were subsequently presented with two hypothetical scenarios: In the good prognosis scenario, a child with medulloblastoma has a good prognosis for survival even with reduced dose of radiation; in the poor prognosis scenario, a child has a poor prognosis for survival even with aggressive therapy (higher dose of radiation). For each scenario participants were asked whether or not they would reduce radiation, what factors would influence their decision, and how certain they would need to be about the child’s prognosis in order to use that information in their decision making. All transcriptions were imported into NVivo9 (QSR) for coding and analysis. Due to the nature of focus group data, the results indicate the relative importance of different ideas/opinions based on how often the ideas were expressed. However, data on the exact number of times an idea was mentioned or the number of people who expressed an idea are not included because people may share an opinion with the group but not voice it or an idea may be expressed multiple times by the same participant. Consequently, data are described in terms of the relative importance or ranking of ideas without numerical information linked to the statements. All participants consented to participate. Ethics approval was received from all participating institutions.

**RESULTS.** Parents and providers showed differences in which factors they believe have the greatest impact on quality of life for children with medulloblastoma and their families. For parents, the most important factor is social functioning and their child’s ability to make friends and have a social life. In contrast, providers thought that parents cared most about their child’s cognitive functioning and ability to attend and perform in school.

**Conclusion.** Understanding parents’ perspectives on quality of life is important in terms of providing support services that target the areas that the parents prioritize. The types of functioning that are most strongly correlated with quality of life from the parents’ perspective may be the ones that should be targeted to protect during treatment.

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RESULTS

A total of 32 people agreed to participate in the study; 16 parents (8% of those approached) and 16 providers (50% of those approached) (Table I). In Toronto, one focus group was held with 12 parents (four families having both parents participate and four families had only one parent present). In Vancouver, four parents from four different families participated in one focus group. The providers represented many roles including oncologists, nurses, speech-language pathologists, social workers, physiotherapists, and clinical and post-doctoral fellows. The parents all had living children who have or had medulloblastoma. The length of time since their children underwent treatment varied among parents; some were just starting treatment, others had completed treatment within the last few years and some had completed treatment many years ago. The severity of their children’s treatment-related deficits varied widely from moderate to severe.

The following is a descriptive analysis of the opinions and perspectives expressed in the focus groups and interviews.

QUALITY OF LIFE

Parents and providers were asked which side effects of therapy most impacted the quality of life of their child and the family. Six factors were mentioned by parents and five factors by providers as key to quality of life (Table II).

SOCIAL FUNCTIONING

For parents, the most devastating consequence of treatment is the effect on their child’s ability to socialize. Parents describe their child as more immature than peers and speaking slowly such that it becomes difficult to engage in conversations with a natural flow, both of which contribute to social challenges.

Providers did not identify the social consequences as a primary factor affecting quality of life except within the context of social relations being an element of independence.

INDEPENDENCE

Parents talked about their child becoming independent as pivotal to quality of life. They describe wanting their children to live on their own and support themselves.

TABLE I. Participant Demographics

<table>
<thead>
<tr>
<th># of parents</th>
<th>16</th>
</tr>
</thead>
<tbody>
<tr>
<td>Avg. age of child at diagnosis</td>
<td>7.4</td>
</tr>
<tr>
<td>Min. age of child at diagnosis</td>
<td>3.5</td>
</tr>
<tr>
<td>Max. age of child at diagnosis</td>
<td>14.5</td>
</tr>
<tr>
<td>Avg. # years between diagnosis and study participation</td>
<td>10</td>
</tr>
<tr>
<td>Min. # years between diagnosis and study participation</td>
<td>5.4</td>
</tr>
<tr>
<td>Max. # years between diagnosis and study participation</td>
<td>15.9</td>
</tr>
<tr>
<td># of providers</td>
<td>16</td>
</tr>
<tr>
<td>Nurses</td>
<td>5</td>
</tr>
<tr>
<td>Speech-language pathologists</td>
<td>1</td>
</tr>
<tr>
<td>Social workers</td>
<td>1</td>
</tr>
<tr>
<td>Physiotherapists</td>
<td>2</td>
</tr>
<tr>
<td>Clinical fellow/post-doctoral fellow</td>
<td>2</td>
</tr>
<tr>
<td>Physicians (neurosurgeons/pediatric oncologists)</td>
<td>5</td>
</tr>
<tr>
<td>Total # of participants</td>
<td>32</td>
</tr>
</tbody>
</table>

TABLE II. Most Important Factors Impacting Quality of Life for Children With Medulloblastoma and Their Families

<table>
<thead>
<tr>
<th>Parents</th>
<th>Providers</th>
</tr>
</thead>
<tbody>
<tr>
<td>2. Independence [3][19%]</td>
<td>2. Independence [4][25%]</td>
</tr>
<tr>
<td>5. Physical functioning [2][12.5%]</td>
<td>5. Social functioning [0][0%]</td>
</tr>
<tr>
<td>6. Pain [1][6.3%]</td>
<td>6. Physical functioning [0][0%]</td>
</tr>
<tr>
<td>7. Financial burden [0][0%]</td>
<td>7. Financial burden [0][0%]</td>
</tr>
</tbody>
</table>

Number and percentage of participants who mentioned the factor indicated in [ ].

Providers thought that independence was very important to parents; more so than the parents expressed. In addition to describing financial independence and ability to live on their own, providers also mentioned physical independence such as being able to feed themselves and walk.

COGNITIVE FUNCTIONING

For two parents, the deterioration of cognitive functioning had a devastating effect on quality of life. Providers largely agreed that cognition is of paramount importance to parents and thought this was the most important factor in quality of life for parents.

FAMILY FUNCTIONING

Only one parent described her healthy child’s resentment of the focus and attention directed towards the sick child and how this has hindered the interactions within the family. In general, parents did not view family functioning as a significant issue. In contrast, providers thought that family functioning played a larger role in quality of life. They described the chronic suffering and disruption of families.

PAIN, PHYSICAL, AND FINANCIAL BURDENS

Pain, physical functioning, and financial burdens were rarely mentioned by parents or providers.

QUALITY OF LIFE VERSUS SURVIVORSHIP

The issue of quality of life versus survival surfaced in various contexts. Parents and providers tended to diverge between survival at all costs or quality of life before longevity.

SURVIVAL AT ALL COSTS

Most parents said that they would want their child to have aggressive treatment regardless of the impact on quality of life. For these parents, having their child alive was the most important thing. This sentiment was reflected in comments such as:

I would rather be as aggressive as possible, probably very selfishly, so that I can make sure she lives and I will do my best to...
build on her strengths so she develops and becomes the best she can be. I wouldn’t trade her life for fewer side effects.

[Parent]

A few providers said that they make treatment decisions based primarily on keeping the child alive and that quality of life is secondary to survival. However, many providers who said that they make treatment decisions that prioritize survival over quality of life said that they do so because that is what they believe parents prefer.

QUALITY OF LIFE OVER LONGEVITY

A minority of parents and providers emphasized the importance of factoring quality of life into treatment decisions rather than automatically striving for survival. Parents expressed that knowing what they know now about how treatment affects quality of life, they would have chosen less aggressive therapy at the risk of their child dying.

The providers who prioritize quality of life explained that when they make treatment recommendations they factor in quality of life and do not necessarily recommend to parents the treatment with the greatest odds of survival.

GOOD PROGNOSIS SCENARIO

Factors Affecting Decision About Radiation

Parents and providers identified several factors that would play into their decision as to whether or not to reduce or eliminate radiation in the event of a good prognosis.

Parents. For parents the most important factor in deciding whether or not to reduce/eliminate radiation is how this reduction would affect the child’s probability of survival. Most parents would be unwilling to accept any decrease in likelihood of survival regardless of what benefits could result from less exposure to radiation. Parents were also generally unwilling to accept any increased risk of recurrence.

Most parents explained that the decision to reduce radiation would have to be based on your child’s odds of surviving with less aggressive treatment and the improvement in your child’s quality of life.

A minority of parents would accept a reduced likelihood of survival and an increased risk of recurrence if serious side effects could be avoided. A few parents would base their decision on treatment options and probability of survival in the event of recurrence.

Providers. Providers mentioned different clinical factors as determinants in how much, if any, radiation they would use. For instance, whether surgery completely removed the tumor or if there is residual disease would influence the use of radiation; with residual, they may be less inclined to reduce radiation. Contrariwise, any complications from surgery may lead to less aggressive radiation. The child’s age and functioning/current deficits would also have an impact on the radiation used. The older the child the less likely the providers would decrease the dose, on the other hand if the child already has significant deficits then they may be more likely to decrease radiation. As well, a good prognostic based on a genetic test would weigh on the decision of how much radiation would be used.

Some providers indicated that their decision about the use of radiation would take into account the family’s preferences and whether the family prioritizes survival or quality of life. As with the parents, for a few providers treatment options in the event of relapse would be a significant factor in the decision about use of radiation.

Certainty of Prognosis

Parents and providers would need to be very confident that their child was correctly categorized as having a good prognosis before they would be willing to decrease radiation. Parents made comments like needing to be “very sure” or “100%” certain that their child really did have a good prognosis. All providers (except one) said the test would need to be at least 90% accurate, with some requiring accuracy of 95% or 99%. There was significant variation in the acceptable likelihood of survival without radiation in order to feel comfortable reducing radiation for the potential gains in quality of life. Acceptable rates of survival ranged from 55% to 70% with no radiation to 90% with some radiation.

POOR PROGNOSIS SCENARIO

Factors Affecting Decision About Radiation

Parents and providers identified factors that would influence their decision as to whether or not to reduce or eliminate radiation in the event of a poor prognosis.

Parents. Most parents were generally unwilling to reduce radiation in the event of a poor prognosis because, regardless of the overall odds for children with a poor prognosis, for any individual child there is always a chance that the child will survive. In contrast, some parents would reduce radiation to improve quality of life even if it meant the child would die sooner.

Providers. The main factor that would influence the providers’ decision about reducing radiation is the likelihood that the child would survive with aggressive treatment. The cut-off varied but in general providers would not eliminate radiation unless the child’s odds of survival with radiation were below 5–10%.

Several providers stipulated that they would only use less aggressive therapy if radiation was going to be futile and they could provide them with an alternative experimental treatment. These providers made two arguments for the use of experimental treatment. First, it’s possible that some treatment may work for these poor prognosis children and it would be wrong to write them off when it’s possible that a treatment exists that could cure them. As one provider explained, the value of a biomarker that identifies children with a poor prognosis is identifying patients with whom to use experimental therapies. Second, unless we test experimental therapies on poor prognosis children scientists will never be able to develop a cure for these patients.

Certainty of Prognosis

Providers and parents agreed that the accuracy of the biomarker test would need to be very high. As one parent put it, the accuracy would need to be “pretty damn good.” Providers generally wanted accuracy over 90% with some saying they would need accuracy of 95–98%.

Who Should Make Treatment Decisions?

Although the focus groups asked parents and providers what would influence their use of less aggressive therapy, both types of participants raised the issue of whether or not parents should be
making this decision at all. Overall, they felt that this decision should be left to the doctors because they have the medical knowledge to make an informed decision and because parents are not emotionally able to make this decision. Parents and providers described how unprepared parents would be to make the decision:

Parents and providers also explained that doctors are better qualified to make the decision about whether or not treatment intensity is reduced because parents do not understand the implications of radiation and the reality of how much quality of life can be affected by it.

Parents also expressed that they may feel tremendous guilt if they opt for reduced radiation and the child does not survive. They would find it easier to cope with if the decision had been made by a doctor. Although it was a minority position, a couple of parents did state that they would like to work with the doctor on making the decision about treatment intensity.

**DISCUSSION**

Parents and providers showed differences in which factors they believe have the greatest impact on quality of life for children with medulloblastoma and their families. For parents, the most important factor is social functioning and their child’s ability to make friends and have a social life. In contrast, providers thought that parents cared most about their child’s cognitive functioning and ability to attend and perform in school. Understanding parents’ perspectives on quality of life is important in terms of providing support services that target the areas that the parents prioritize. It may also be the case that different aspects of therapy differentially affect different types of functioning. If so, the types of functioning that are most strongly correlated with quality of life from the parents’ perspective may be the ones that should be targeted to protect during treatment, if that option exists. It is necessary to further determine which factors of quality of life matter most to parents so as to better inform potential treatment ramifications.

Finally, it may be that there is only an apparent discrepancy between parents and providers perspectives, and their comments actually reflect similar concerns, but the concerns are manifest differently. Parents have the most opportunity to observe their children in social situations—and as such the thinking difficulties children experience (i.e., appearing immature) are manifest as social problems for parents. Providers on the other hand are much more likely to use information from standardized cognitive assessments and interact with school personnel than observe children socially, and hence they focus on the cognitive aspects of children’s thinking problems.

In the 1970s and early 80s, radiation was used upfront for many young children. In the late 1980s, agreement was reached to try to avoid radiation in this population. This was mainly driven by the understanding that quality of survival was poor when radiation was given in this young population [14]. This decision was made at the potential cost of a lower survival. In older children, the main focus during the last 20 years has been to improve survival rates, but even in this age group attempts have been made to reduce craniospinal radiation. Now, we are moving in a new era, as molecular biology has allowed us to identify subgroup of patients with different outcomes. With this in mind, the question is whether care providers and parents are ready to use this information to tailor treatment choices.

Based on discussions with parents and providers, it is unclear who should be involved in making these decisions. Many parents recognize that they would be unable or unwilling to make a decision that could result in decreasing their child’s odds of survival either because they lack the knowledge to make an informed decision, are too stressed and emotionally overwhelmed at the time the decision needs to get made, or would feel too much responsibility for their child’s death if the child did not survive. These parents would prefer that the doctors make the decision about the most appropriate treatment for their child. However, many of the providers who participated in the study indicated that they think it is appropriate to make the decision in consultation with parents and believe that parents should have a say in whether quality of life or survival is prioritized in treatment and that they already take into account what is explicitly or perceived to be the parents’ preferences. The problem with providers taking parent preferences into account is that parents cannot imagine what quality of life will be like for their child post-treatment so the parents would be contributing to the conversation without the experience and full understanding of the long-term situation that would enable them to formulate a truly informed opinion.

Additionally, it is important to address how quality of life and the long-term outcomes can be integrated into treatment protocols. This issue is particularly relevant given that parents differ in how they prioritize quality of life. As well, if protocols are intended to be used across institutions it needs to be considered whether different beliefs and values about survival versus quality of life vary across institutions and, if so, whether these differences should be factored into how the protocols are used.

The ethics surrounding the decision making process for treatment options remain unclear. Even if there might be no permanent ethical answer, future discussions should consider how and to what extent parents’ opinions can be integrated into the decision making process.

As with any focus group study, results are not generalizable. Generalizability may be further impacted by the sample size. As well, all sessions were conducted in Canada and the differences between the Canadian and other healthcare systems may lead to differences in priorities and practices in treatment. Only parents of children currently undergoing treatment or who survived treatment participated; their perspectives may differ from parents whose children did not survive, as well the time that has elapsed after the end of treatment. Also, the perspective of the retrospective situation of parents after the end of treatment is different from the prospective situation of the parents discussing the prognostic information at time of diagnosis. The experience of the caregiver might have an influence on parents’ perspectives. Including former patients would have been of interest. There is considerable evidence [15] that the perspectives of parents, teachers, child, and peers vary dramatically when using standardized measures to evaluate psycho-social functioning—and such differences in perspective may also be evident when information is gleaned via focus group. As such, our results have to be considered in the context of which they were obtained and that the source of information (the parents) may not reflect the opinions or preferences of other sources (peers, or the affected individual) had we sought them.

**REFERENCES**


