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Treating the Pediatric Cancer Patient: Insights that have stood the test of time

A Commentary

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Funding: This work is supported, in part, by the Intramural Research Programs of the NIH, National Cancer Institute, Center for Cancer Research.

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In 1977, Gogan, O'Malley & Foster published a critically important and insightful review of 75 papers spanning 1944-1976 addressing the psychological and social issues affecting children with cancer. In some ways, a great deal has changed over the past 42 years. In other ways, issues needing the attention of those involved in the treatment of seriously ill children remain the same today.

Describing patient and family experiences, Gogan and colleagues addressed the “trauma” that can impact a child’s ability to cope, particularly early in their treatment. Today, while the whole family still experiences significant distress associated with a cancer diagnosis, we have learned that most children and parents cope well and exhibit resilience (Kazak & Noll, 2015; Phipps, et al., 2015; Okado, Rowley, Schepers, Long, & Phipps, 2018). With targeted social support, clinical intervention, and education, they can experience growth during the cancer trajectory (Turner, Hutchinson, & Wilson, 2018). To identify challenges early, evidence-based approaches to screening are becoming an essential step in the delivery of psychosocial care (Kazak et al., 2017, 2018). Patient-reported outcomes (PROs) are increasingly being used in clinical practice and can enhance identification of health-related quality of life problems and satisfaction with care in children with cancer (Schepers, Haverman, Zadeh, Grootenhuis, & Wiener, 2016, Schepers et al., 2017).

At the time this review was published, children with cancer were beginning to live “many months or years”. Gogan and colleagues recognized that with increased survivorship attention must “extend beyond treatment of the malignancy” and be given to quality of life and improving communication among the patient, family and health care professional. With significant improvements in the long term-survival of children with cancer, these same issues remain as relevant today as they were 4 decades ago. Thus, a focus today is on alleviating suffering from

the time of diagnosis through survivorship in order to improve short- and long-term outcomes. Pediatric palliative care, which addresses many of the physical, emotional and spiritual challenges children with cancer face regardless of disease outcome, didn't exist at the time this paper was published (Snaman, McCarthy, Wiener, & Wolfe, in press).

With increased survival has come new challenges that weren't anticipated in the 1970's. Sixty percent of children who survive cancer suffer a range of often devastating late effects such as secondary cancers and infertility as well as psychosocial difficulties not related to physical health problems (Brier et al., 2011; Brinkman et al, 2013, 2014; Ford et al, 2014; Kunin-Batson et al., 2016; Lown, Phillips, Schwartz, Rosenberg, & Jones, 2015; Wiener, Battles, Bernstein, Long, Mansky, Mackall, 2006). With approximately 375,00 adult survivors of childhood cancer in the US, this is a significant issue. The Children's Oncology Group (COG) has outlined guidelines that recommend regular surveillance and care for those who have completed cancer therapy (COG, 2018). Nevertheless, many childhood cancer survivors still do not receive appropriate long-term follow-up care.

Gogan and colleagues devoted an entire section of their review to the issue of what we today call prognostic disclosure to seriously ill children. They addressed the transition from a "protective approach" (a style of communication that shielded children from the harms of bad news) to a more "open approach" (discussing the possibility of death with a child). In fact, by the late 1970's, the recommended approach to prognostic disclosure in pediatrics shifted largely from "never tell" to "always tell." This was in part due to research which showed that even young children with cancer come to understand their prognosis even if they are not told (Waechter, 1971; Spinetta, 1974; Bluebond-Langner, 1974, 1978). Without an opportunity to discuss this knowledge, it was thought, knowing their prognosis was distressing for children

(Slavin, O'Malley, Koocher & Foster 1982). In the decades following, these psychological reasons for open discussion were supplemented by developments in bioethics: an increasing rejection of paternalism and emphasis on the developing autonomy of children. We now have advance care planning guides that provide youth an opportunity to document specific preferences for their care and to share with their loved ones how they would like to be remembered if cure is not possible (Zadeh, Pao & Wiener, 2015). This is a far cry from the earlier approach that advised parents to cheerfully assure the child that he or she will soon be well (Evans, 1968; Toch, 1964).

Yet, today we appreciate that disclosure is not a black and white issue. Children and adolescents, like adults, have varying preferences for what sort of information they want, when they want to hear it, and from whom (Bluebond-Langner, Belasco, & Wander 2010; Day, Jones, Langner, & Bluebond-Langner, 2016; Kelly, Mowbray, Pyke Grimm & Hinds 2016; Sisk, Mack, Wiener, Bluebond-Langner, & Wolfe, 2016; Weaver, Baker, Gattuso, Gibson, Sykes & Hinds 2015). Enforcing openness can be harmful to the relationship between parents, child and clinicians (Rosenberg, Wolfe, Wiener, Lyon, & Feudtner, 2016).

The review by Gogan and colleagues showed sensitivity to the choice of concepts and methods used to understand the impact of cancer on children and their families. The authors moved away from earlier psycho-dynamic theories and began to view patterns of behavior as efforts to cope with the impact of cancer - noting that there is no generally accepted definition of "coping". This review was one of the first review papers that went beyond the focus on "parent-child interaction to a more complex "system approach," addressing the needs of siblings. While siblings' long-term psychosocial adjustment to childhood cancer is still poorly understood, there has been considerable data supporting a strong need for sibling support (Sahler, O. J et al., 1996;

Long, Lehmann, Gerhardt, Carpenter, Marsland, & Alderfer, 2018; Gerhardt, Lehmann, Long, & Alderfer, 2015). We continue to see that cancer is a complex phenomenon which requires continuous reflection on the adequacy of the frameworks which we use in our research and by extension practice (Kupst & Patenaude, 2015).

In summary, “Treating the Pediatric Cancer Patient” by Gogan, O’Malley & Foster (1977) deserves to be acknowledged as having historical and prescient significance. Over the past 4 decades, many changes have emerged. Health related groups of psychosocial professionals have been developed and have thrived over the years. The open approach Gogan and colleagues spoke about is common practice today. The move away from the physician as the expert and the importance of incorporating the whole family in the hospital system can’t be understated. This includes the engagement of parents as partners in care and shared decision-making with the child. And many of the issues described that were based on limited objective data are now supported in evidence-based standards for pediatric psychosocial care of children with cancer and their families (Wiener, Kazak, Noll, Patenaude, & Kupst, 2015).

We are once again in a time of innovation in treatment. Today we have clinical trials, personalized medicine, fertility/cryo-preservation, proton beam therapy, and treatment with immunotherapy as well as cancer predisposition clinics. There is rapid development of E/mHealth technologies and the use of telemedicine. We need to carefully anticipate new practical, clinical and ethical challenges that may arise and how these changes can affect our approaches to care. Nevertheless, empirical studies are still needed on the 7 areas which Gogan and colleagues propitiously list in their article: outcomes of open communication, physician communication, family coping strategies, whole family therapies, intervention evaluation, survivorship, and the reorganization of care. Ideally, a stronger evidence base will inform the

development of interventions that improve communication, bolster the clinician-parent-patient relationship, and enhance the quality of life of children and their family members from the time of diagnosis, through survivorship or end of life and into bereavement.

References

- Bluebond-Langner, M. (1974). I Know, Do You? A Study of Awareness, Communication and Coping in Terminally Ill Children. In Anticipatory Grief. Schoenberg, B., Carr, A.C., Peretz, D. Kutscher, A.H. & Goldberg, I.K. (eds). Columbia University Press, New York.
- Bluebond-Langner, M. (1978). *The Private Worlds of Dying Children*. Princeton, NJ: Princeton University Press.
- Bluebond-Langner, M., Belasco, J., & Wander M. (2010). Involving Children In Decisions About Their Care and Treatment. *Nursing Clinics of North America*, 45 (3), 329-345.
- Brier, M. J., Kazak, A. E., Derosa, B. W., Hocking, M. C., Schwartz, L. A., Ginsverg, J. P. ... Ittenbach, R. F. (2011). Profiles of Health Competence Beliefs Among Young Adult Survivors of Childhood Cancer. *Journal of Adolescent and Young Adult Oncology*, 1(4), 195–202.
- Brinkman, T. M., Zhang, N., Recklitis, C.J., Kimberg, C., Zeltzer, L. K., Muriel, A. C., ...Krull, K.R. (2014). Suicide ideation and associated mortality in adult survivors of childhood cancer. *Cancer*, 120(2), 271–277.
- Brinkman, T. M., Zhang, N., Ullrich, N. J., Brouwers, P., Green, D. M., Srivastava, D. K., Zeltzer, L. K., Stovall, M., Robison, L. L., ... Krull, K. R. (2012). Psychoactive medication use and neurocognitive function in adult survivors of childhood cancer: a report from the Childhood Cancer Survivor study. *Pediatric blood & cancer*, 60(3), 486-93.
- Children’s Oncology Group. Long-Term Follow-Up Guidelines, Version 5.0 – October, 2018. http://www.survivorshipguidelines.org/pdf/2018/COG_LTFU_Guidelines_v5.pdf
- Day, E., Jones, L., Langner, R., & Bluebond-Langner, M. (2016). Current understanding of decision-making in adolescents with cancer: a narrative systematic review”. *Palliative Medicine*, 30(10), 920-34.
- Evans, A. E. (1968). If a child must die. *New England Journal of Medicine*, 273, 138-142.
- Ford, J. S., Kawashima, T., Whitton, J., Leisenring, W. Laverdière C, Stovall, M.,Sklar, C. A. (2014). Psychosexual functioning among adult female survivors of childhood cancer: a report from the childhood cancer survivor study. *Journal of Clinical Oncology*, 32(28), 3126–3136.
- Gerhardt, C. A., Lehmann, V., Long, K.A., Alderfer, M.A. (2015). Supporting siblings as a standard of care in pediatric oncology. *Pediatric Blood & Cancer*, 62 Suppl 5, S750-804.

Gogan, J. L., O'Malley J. E., Foster, D. J. (1977). Treating the pediatric cancer patient: A review. *Journal of Pediatric Psychology*, 2(2), 42-48.

Kazak, A. E, Hwang, W. T., Chen, F. F., Askins, M.A., Carlson, O., Argueta-Ortiz, F., Barakat, L. P. (2018). Screening for family psychosocial risk in pediatric cancer: Validation of the Psychosocial Assessment Tool (PAT) Version 3. *Journal of Pediatric Psychology*, 43(7), 737-748.

Kazak, A. E. & Noll, R. B. (2015) The integration of psychology in pediatric oncology research and practice. *American Psychologist*, 70(2), 146-158.

Kazak, A. E., Barakat, L. P., Askins, M. A., McCafferty, M., Lattomus, A., Ruppe, N., Deatrck, J. (2017). Provider Perspectives on the Implementation of Psychosocial Risk Screening in Pediatric Cancer. *Journal of Pediatric Psychology*, 42(6), 700-710.

Kelly, K. P., Mowbray, C., Pyke-Grimm, K. & Hinds, P.S. (2017) Identifying a conceptual shift in child and adolescent reported treatment decision making: "Having a say, as I need at the time". *Pediatric Blood and Cancer*, 64(4), 1-8.

Kunin-Batson, A. S., Lu, X., Balsamo, L., Graber, K., Devidas, M., Hunger, S. P., Carroll, W. L., Winick, N. J., Mattano, L. A., Maloney, K. W., ... Kadan-Lottick, N. S. (2016). Prevalence and predictors of anxiety and depression after completion of chemotherapy for childhood acute lymphoblastic leukemia: A prospective longitudinal study. *Cancer*, 122(10), 1608-17.

Kupst, M J & Patenaude, A. F. Coping and adaptation in pediatric cancer: Current perspectives. In: *Pediatric Psychosocial Oncology: Textbook for Multidisciplinary Care*. Springer, 67-80.

Long, K. A., Lehmann, V., Gerhardt, C. A., Carpenter, A. L., Marsland, A. L., Alderfer, M.A. (2018). Psychosocial functioning and risk factors among siblings of children with cancer. An updated systematic review. *Psychooncology*, 27(6),1467-1479.

Lown, E. A., Phillips, F., Schwartz, L. A., Rosenberg, A. R., & Jones, B. (2015). Psychosocial Follow-Up in Survivorship as a Standard of Care in Pediatric Oncology. *Pediatric blood & cancer*, 62 Suppl 5(Suppl 5), S514-84.

Okado, Y., Rowley, C., Schepers, S. A., Long, A. M., Phipps, S. (2018). Profiles of adjustment in pediatric cancer survivors and their prediction by earlier psychosocial factors. *Journal of Pediatric Psychology*, 43(9):1047-1058.

Phillips, S. M., Padgett, L. S., Leisenring, W. M., Stratton, K. K., Bishop, K., Krull, K. R., et al. (2015). Survivors of childhood cancer in the United States: Prevalence and burden of morbidity. *Cancer Epidemiol Biomarkers Prev*, 24(4), 653-663.

Phipps, S., Long, A., Willard, V. W., Okado, Y, Hudson, M., Huang, Q., Zhang H., Noll, R. (2015). Parents of children with cancer: At-risk or resilient? *Journal of Pediatric Psychology*, 40(9):914-25.

Rosenberg, A. R., Wolfe, J., Wiener, L., Lyon, M., Feudtner, C. (2016). Ethics, emotions, and the skills of talking about progressive disease with terminally ill adolescents: a narrative review. *JAMA Pediatrics*, 170(12):1216-1223.

Sahler, O. J., Roghmann, K. J., Mulhern, R. K., Carpenter, P. J., Sargent, J. R., Copeland, D. R., ...Dolgin, M. J. (1997). Sibling Adaptation to childhood cancer collaborative study: the association of sibling adaptation with maternal well-being, physical health, and resource use. *J Dev Behav Pediatr*, 18(4), 233-43.

Schepers, S. A., Sint Nicolaas, S. M., Haverman, L., Wensing, M., van Meerteren, A. Y. N., Veening, M.A..... Grootenhuis, M. (2017) Real-world implementation of electronic patient-reported outcomes in outpatient pediatric cancer care. *Psycho-Oncology*, 26, 951–959.

Schepers, S. A., Haverman, L., Zadeh, S., Grootenhuis, M. A., & Wiener, L. (2016). Healthcare Professionals' Preferences and Perceived Barriers for Routine Assessment of Patient-Reported Outcomes in Pediatric Oncology Practice: Moving Toward International Processes of Change. *Pediatric Blood & Cancer*, 63(12), 2181-2188.

Sisk, B. A., Bluebond-Langner, M., Mack, J., Wiener, L., Wolfe, J. (2016). Prognostic disclosure to children: Truth telling in the past, present and future. *Pediatrics*, 138 (3), e2 016127826634.

Slavin, L.A., O'Malley, J.E., Koocher, G.P. & Foster, D.J. (1982). Communication of the Cancer Diagnosis to Pediatric Patients: Impact of Long-Term Adjustment. *American Journal of Psychiatry*, 139(2), 179-183.

Snaman, J. M., McCarthy, S., Wiener, L. Wolfe, J. Pediatric palliative care in oncology: It's all in the family. *JCO*, *in press*.

Spinetta, J. (1974). The dying child's awareness of death. *Psychological Bulletin*, 81(4), 256-260.

Toch, R. (1964). Management of the child with a fatal disease. *Clinical Pediatrics*, 3, 418-427.

Turner J. K., Hutchinson, A., Wilson, C. (2018). Correlates of post-traumatic growth following childhood and adolescent cancer: A systematic review and meta-analysis. *Psychooncology*, 27(4), 1100-1109.

Waechter, E. H. (1971). Children's awareness of fatal illness. *The American Journal of Nursing*, 71 (6), 1168-1172.

Weaver, M.S., Baker, J.N., Gattuso, J.S., Gibson, D.V., Sykes, A.D. & Hinds, P.S. (2015). Adolescents' Preferences for Treatment Decisional Involvement During Their Cancer. *Cancer*, 121(24), 4416-4424.

Wiener, L., Kazak, A. E., Noll, R., Patenaude, A. F., Kupst, MJ. (2015). Standards for the Psychosocial Care of Children with Cancer and Their Families. *Pediatric Blood and Cancer*, 62 (S5), 425.

Wiener, L., Battles, H. B., Bernstein, D., Long, L., Mansky, P., Mackall, C. (2006). Persistent Psychological Distress in Long-Term Survivors of Pediatric Sarcoma, *Psycho-Oncology* 15, 898-910.

Zadeh, S., Pao, M., & Wiener, L. Opening end-of life discussions: How to introduce Voicing My CHOICES™, an advance care planning guide for adolescents and young adults. (2015). *Palliative and Supportive Care*, 13 (3), 591-599.