Institution: University of Sheffield

Unit of Assessment: 4 – Psychology, Psychiatry and Neuroscience

Title of case study: Improving quality of life among children with cancer: Impact on clinical guidelines and education of children and families

1. Summary of the impact

Cancer treatment for children is one of the success stories of medical care in the twentieth century. Survival increased from almost zero in the 1950s to today, when treatment for some child cancers results in over 90% survival. These improved survival rates have, however, been achieved through use of highly aggressive treatment protocols, with adverse implications for the child's cognitive, emotional and social development and the burden of care on families. Nationally, researchers at Sheffield were among the first to identify the extent to which children continued to show psychological and behavioural problems, even long after the end of treatment. As such, they contributed significantly to discussions about how to balance medical treatment to control the cancer while taking into account the immediate and longer-term impacts on child quality of life and parents' psychological well-being. The work has had direct implications for both national and international clinical guidelines, and assessment of quality of life in national clinical trials. It has also resulted in user-friendly information for schools and families.

2. Underpinning research.

Context

Cancer in childhood affects approximately 1 in 600 children under 15 years, with around 1,400 new cases per year in the UK. Cancer includes a range of conditions varying in severity, responsiveness to treatment and impact on quality of life. After diagnosis, children are treated with a combination of radiotherapy, chemotherapy and surgery. Chemotherapy is continued for a finite period of time depending on the specific cancer (the most common, acute lymphoblastic leukaemia (ALL), involves treatment for two years for girls and three for boys). Although children are considered 'cured,' relapse remains possible. The principle guiding current care is to achieve a balance between the toxicity of prescribed treatment with optimal long-term well-being.

Diagnosis and treatment

On diagnosis, parents need to understand the life-threatening nature of the disease, the very complex treatment protocols and take responsibility for day-to-day treatment (administering medication, monitoring the child's general health). For many cancers, the average age on diagnosis is below five years, creating challenges in terms of communication and facilitating child cooperation with, and adherence to, treatment.

Work in Sheffield led by Professor Christine Eiser, funded by the Health Technology Assessment Programme, showed that children experienced many difficulties in school, aggravated by changes in their appearance following chemotherapy [**R1**]. Children missed a large amount of school time and fell behind in schoolwork. They could be teased by others, and teachers were not always sympathetic, partly because they had insufficient understanding of the disease and treatment. Parents described their difficulties trying to give the child a normal life while also ensuring they were not exposed to infection. Many 'spoiled' the child to compensate for a limited quality of life (QoL), and pre-school children posed special challenges because of poorly developed language skills. Extensive work was conducted concerned with measurement of QoL and relationship between parent and child ratings [**R2**].

Survivors

Survival rates in childhood cancer have increased in recent years, so that approximately 75% can expect five-year event free survival. However, two-thirds experience one or more late-effects, varying from relatively benign to severe and debilitating. Consequently, children are recommended to attend follow-up for some years after the end of treatment.

Increasing survival rates and the delayed appearance of some late-effects (e.g. infertility) pose organisational challenges for follow-up. Children are treated in paediatric departments, but longer-term surveillance requires attendance in adult hospitals, with the result that some are lost to specialist care.

In response, detailed recommendations regarding surveillance and treatment of late-effects have been reported by working groups in the US and UK. One solution proposed by a UK multidisciplinary Working Party including Eiser [**R3**] was to describe a three-level model of care, where surveillance was matched to likelihood of late-effects.

In Sheffield, Eiser's research reported an initial evaluation of this model [**R4**], and confirmed that staff were reliably able to categorise children in terms of this model; i.e. they agreed on the level of care most appropriate for hypothetical cases differing in terms of diagnosis, treatment and chronological age. The researchers also sought the views of cancer survivors themselves about the kind of care they would prefer [**R5**].

The original concern was about physical late-effects experienced by survivors (e.g. endocrine, cardiac and fertility implications), but the research showed that there were also psychological consequences. The incidence of PTSD was 13.9%, which appears high but was shown to be comparable with figures from the US [**R6**]. It was concluded that decisions to discharge survivors from care must take account of psychological as well as physical late-effects and recommended routine screening for psychological late-effects.

3. References to the research

R1 Eiser, C., Morse, R. (2001). Quality of life measures in chronic diseases of childhood. *Health Technology Assessment*, 5 (4). (Citations: 259)

R2 Clarke, S.A., Davies, H., Jenney, M., Glaser, A., Eiser, C. (2005). Parental communication and children's behaviour following diagnosis of childhood leukaemia. *Psycho-Oncology,* 4: 274-81. doi: 10.1002/pon.843 (Citations: 16)

R3 Wallace, W.H., Blacklay, A., Eiser, C., Davies, H., Hawkins, M., Levitt, G.A., Jenney, M.E., Late Effects Committee of the United Kingdom Children's Cancer Study Group (UKCCSG) (2001). Developing strategies for long term follow up of survivors of childhood cancer. *BMJ*, 323 (7307), 271-4. doi: 10.1136/bmj.323.7307.271 (Citations: 101)

R4 Eiser, C., Absolom, K., Greenfield, D. et al. (2006). Follow-up after childhood cancer: Evaluation of a three-level model. *European Journal of Cancer*, 42: 3186-90. doi: 10.1016/j.ejca.2006.08.001 (Citations: 10)

R5 Michel, G., Greenfield, D.M., Absolom, K., Ross, R.J., Davies, H., Eiser, C. (2009). Follow-up care after childhood cancer: survivors' expectations and preferences for care. *Eur. J. Cancer,* 45:1616-23. doi: 10.1016/j.ejca.2009.02.026 (Citations: 9)

R6 Taylor, N., Absolom, K., Snowden, J., Eiser, C. (2011). Need for psychological follow-up among young adult survivors of childhood cancer. *Eur. J. Cancer*, 21: 52-8 doi: 10.1111/j.1365-2354.2011.01281.x (Citations:3)

4. Details of the impact

These new insights into the issues faced by survivors of childhood cancer have implications for the work of practitioners, information available for survivors and their families, and on clinical guidelines in the UK.

Impacts on guidelines for clinicians and other practitioners

The Children's Cancer and Leukaemia Group (CCLG) is responsible for the national organisation of care in the UK, and through a number of Working Groups helps to ensure that children throughout the country receive the highest quality care available. From 1995 to 2005, Eiser was the only psychologist on three Working Groups (Late-effects, Psychosocial, and Palliative care), and chaired the Psychosocial group (1999–2002). The remit was to ensure that treatment and follow-up recognised the quality of life issues associated with cancer treatment. The need for a balanced approach to care was reflected in a series of multidisciplinary meetings in the UK and Europe. The output of these guideline meetings led to the research questions as to whether the current guidelines were acceptable to patients.

To ensure standard care of survivors across the UK, the Late-effects group published guidelines for follow-up of survivors that included recommendations regarding routine assessment for physical and psychological late-effects [**S1**]. Eiser wrote sections on Quality of Life and neuropsychological follow-up. The aims were to make clear to clinicians (including GPs and junior hospital staff) the different ways in which survivors may experience problems and ensure that likely late-effects were systematically investigated. Inclusion of Eiser's sections on quality of life and neuropsychological outcomes was innovative and ensured a continuing debate about achieving balance between quantity and quality of survival. Subsequent guidelines by other national and international groups followed a similar pattern. Sheffield research has been cited in NICE guidelines, originally published in 2005, but still in force today [**S2**], as well as the Scottish equivalent, SIGN, republished in 2013 [**S3**], and CureSearch [**S4**] a non-profit research foundation in the US.

The research has been cited by non-government organisations including UICC (Union for International Cancer Control). Eiser was also asked to write a chapter on the psychosocial aspects of childhood cancer in a UICC report. Eiser has worked closely with cancer charities such as Cancer Research UK, the Lisa Thaxter Trust, CLIC Sargent and ICCCPO (International Confederation of Childhood Cancers). This has involved providing feedback to research meetings, fundraisers and parent self-help groups.

Impact on child education

A good experience at school is considered vital in facilitating normal psychological development and integration into the adult world of work and social relationships. Eiser's work identified problems resulting from teachers' lack of understanding of the disease and reluctance to make demands on children to avoid distressing them. Accurate information and advice about how to manage the child was provided in terms of patient information booklets for parents and teachers, 'Children with a brain tumour in the classroom' and 'Welcome back'. The booklets were distributed to all children's cancer centres in the UK (n=22) and used by clinic nurses as they felt appropriate. Following demand from treatment centres, CCLG has just taken over copyright from CR-UK for 'Welcome back' and 'Children with a brain tumour in the classroom' and the two booklets are being reprinted, combined into one.

Work with survivors showed that some were reluctant to attend follow-up and unclear why they were asked to do so. To address this, the leaflet "What's the point of coming to clinic?' [**S5**] was produced, aimed at children, to explain the purpose behind attending follow-up clinic and the tests that are performed there and to promote the need to adopt a healthy lifestyle. In January 2007, an original print run of 7,000 was made (1,454 remaining: 270 copies were requested in 2012). Due to demand from clinic staff, the booklet is now being reprinted. The pdf was downloaded 114 times in 2012. "What's the point of coming to clinic?" was received positively by young people attending clinic, with 75% learning new information, and greater awareness of risks after cancer treatment and rationale for follow-up. [**S5**]

'After cure' is aimed at young people aged 16+ who have survived cancer. The booklet covers a variety of topics such as follow-up and future care, education, jobs, disability issues, life insurance, mortgages, fertility, travel, survivor groups and useful links, and has been translated into Punjabi, Bengali and Gujarati. [**S6**]

Although there is no specific information on the extent of use in clinics, the CCLG Publications Committee, comprising multi-professional experts in the field of children's cancer (paediatric oncologists, radiographers, nurses and parents) meets four times a year and evaluates publications for usefulness. Only approved publications remain in print.

In line with MRC requirements, a quality of life assessment was included, for the first time, in the most recent trial to determine changes in treatment protocol for children with acute lymphoblastic leukaemia [**S7**]. The basic research conducted by Eiser and Morse [**R1**] contributed significantly to knowledge of how best to measure quality of life in young children and acceptability by medical staff that it could be successfully achieved. Eiser designed the quality of life assessment in the trial and led data collection for the national five-year study. This involved collaboration across 22 treatment centres in the UK and collation of parental questionnaire responses at five time-points during the course of treatment (approximately 900 children involved).

Impacts on public policy

Eiser's group worked with the CLIC Sargent charity to determine the financial impact on families caring for children with cancer. The publication (Eiser, C.; Upton, P. 2007 Costs of caring for a child with cancer: a questionnaire survey, *Child: care, health and development*, 33, 455-9) formed the basis of the charity's 'Cut the red tape' campaign [**S8**]. Following the change from Disability Living allowance (DLA) to Personal Independence Payment (PIP), a memorandum was submitted by CLIC Sargent [**S9**] citing Eiser's findings, that 83% of families incur significant extra costs associated with their child's cancer treatment with 68% of families experiencing worrying financial difficulties following diagnosis. This is coupled with parents often cutting back on working hours to spend more time caring for their child (9/10 parents surveyed do so), leading to decreased income. With other charity partners, CLIC Sargent used the evidence from Eiser's research to successfully persuade the Government not to increase the qualifying period for the new benefit to six months, which would have particularly penalised those with sudden onset conditions such as leukaemia [**S9**].

5. Sources to corroborate the impact

S1 Skinner, R., Wallace, W.H.B., Levitt, G.A. (eds) (2nd edition, 2005). *Therapy based Long-term follow-up. Practice Statement*. UK Children's Cancer Study Group, Late Effects Group. http://www.cclg.org.uk/dynamic_files/LTFU-full.pdf (formerly United Kingdom Children's Cancer Study Group/UK Cancer and Leukaemia Working Party)

S2 NICE: *Improving Outcomes in Children and Young People with Cancer* (http://tinyurl.com/ofd23te) page 26, reference 11 corroborates Eiser's research being used for the guidance.

S3 Scottish Intercollegiate Clinical guidelines, March 2013, *Long term follow up of survivors of childhood cancer: a national clinical guideline*. (Eiser authored sections on Cognitive and psychosocial outcomes, and Long term follow up). http://www.sign.ac.uk/pdf/sign132.pdf, (and references 83, 154 and 217).

S4 Long-term follow up guidelines for survivors of childhood, adolescent and young adult cancers. (2008). CureSearch Children's Oncology Group (http://tinyurl.com/6datav)

S5 *What's the Point of Coming to Clinic?* (http://tinyurl.com/kh8lfe5) corroborates Eiser's authorship of CCLG guidance for children and teenagers.

S6 *After Cure*, Children's Cancer and Leukaemia Group (http://tinyurl.com/kxmppau) page 2 corroborates Eiser's contribution to the guidance

S7 Vora, A., Goulden, N., Wade, R., Mitchell, C., Hancock, R., Rowntree, C., Richards, S. (2013). Treatment reduction for children and young adults with low-risk acute lymphoblastic leukaemia defined by minimal residual disease (UKALL 2003): a randomised controlled trial. *Lancet Oncology*, 14 (3), 199-209. The parallel Quality of Life paper first-authored by Eiser is currently under review.

S8 *Counting the costs of cancer*, CLIC Sargent (http://tinyurl.com/ne5cekc) 2011, Page 5 ref 1 corroborates Eiser's research contribution to the report.

S9 Welfare Reform Bill, Memorandum submitted by CLIC Sargent (WR 46) (http://tinyurl.com/3upcek2) April 2011, Reference 1 corroborates Eiser's research contribution to the memo which allowed CLIC Sargent to contribute