

## **MODELS OF CARE FOR FOLLOW-UP OF CHILDHOOD CANCER SURVIVORS: A SYSTEMATIC REVIEW**

As they grow older, survivors of childhood cancer need access to appropriate expertise within the adult health care system. A wide range of models of care for survivors of cancer have been developed, and reviews have been undertaken focussed primarily upon the effectiveness of alternative approaches in the adult cancer population. The most effective mode of follow-up remains a matter of current debate. This review sought to uncover the existing evidence which underpin modes of follow-up with the aim of influencing clinical practice and identifying key areas and approaches for future research.

The purpose of this evidence briefing is to summarise the findings of the review to inform the Children & Young People Cancer Survivorship Initiative workshop: Providing the Evidence to Achieve Change, London, 24<sup>th</sup> March 2010. The report is to be submitted to the commissioners, Macmillan Cancer Support, and may undergo change during the peer review process. You are therefore asked not to copy, quote, cite, circulate or use this document for any purpose other than to inform the discussion at the workshop.

### **BACKGROUND**

#### **Childhood Cancer and Survival**

Childhood cancer is very rare; nevertheless, in the UK cancer accounts for approximately 20% of all deaths between the ages of 1 and 14 years and each year approximately 1,400 new cases of childhood cancer are diagnosed.<sup>1,2</sup>

Childhood cancers differ in distribution according to age at diagnosis; occurrence is approximately 20% higher in boys than in girls.<sup>1</sup> Acute lymphoblastic leukaemia accounts for a quarter of all childhood cancers.

Lymphomas, brain and spinal tumours, embryonal tumours, bone tumours, soft tissue sarcomas, germ cell and gonadal tumours and carcinoma and melanoma are also seen in children.

There is no evidence that the incidence of childhood cancer has changed substantially in recent years, however improvements in the precision of diagnosis, therapy and supportive care has led to increasing survival rates since the 1960's. Little is known about the long-term consequences of therapy and the need for long-term surveillance has been identified in order to better characterise late-effects specific to this group.<sup>3</sup>

#### **Late effects of Childhood Cancer**

There is a need to balance the risk of late adverse effects against potential gains in duration of survival. After childhood cancer 60% of people who are more than five years from completion of therapy will experience at least one or more treatment- or disease-related late effects of therapy; over 30% of these problems are moderate or severe.<sup>4</sup> Physical, mental or social aspects of health may be affected which may interfere with survivors' autonomy.<sup>5</sup>

#### **Follow-up**

There is general recognition that all survivors of childhood cancer should be followed up for life.<sup>6-8</sup> However, there remains a lack of consensus regarding the optimal setting and strategy for doing this.<sup>7</sup>

The most common method of follow-up in the UK appears to be a cancer centre-based approach with a strong emphasis on paediatric oncology.<sup>9</sup> Other potential models include; specially trained nurses conducting long-term follow-up; transition models for transfer of care to a more age-appropriate provider at a defined age,<sup>9,10</sup> community care based in primary care settings; oncology care

for adults in hospital-based clinics; and patient-driven follow-up (whereby the survivor is responsible for seeking medical care when particular symptoms are present).<sup>9</sup>

## **NATURE OF THE EVIDENCE**

No existing reviews have attempted to compare the effectiveness of different modes of follow-up specifically in survivors of childhood cancer.

A recent systematic review by Aslett et al.<sup>11</sup> investigated current long-term follow-up practices for survivors of childhood cancer. They found that there were a variety of models of care utilised, a lack of consistency in practice, and variation in the level and degree to which long-term survivors were followed up.

Two other reviews were identified, one examined differences between primary and secondary care and the other examined patients' and health care professionals' views about cancer follow-up.<sup>12,13</sup> Both focused on adult cancer survivors.

This review sought to uncover the existing evidence which underpins the following approaches:

- a comparison of alternative communication modalities to face-to-face clinic visits (for example telephone, postal, email or SMS/text-based)
- the use of physician- versus nurse-led follow-up
- the value of hospital staff versus primary care staff to provide clinical contacts

The review was carried out at the Centre for Reviews and Dissemination (CRD) using the methods set out in CRD's guidance on undertaking reviews of effectiveness and NICE public health guidance.<sup>14,15</sup> Full details of the methods will be presented in the final report.

## **THE EVIDENCE**

No controlled studies evaluating methods of providing follow-up care for survivors of childhood cancer were identified. However, observational, single arm or audit-style studies that met our other inclusion criteria, were

identified and collated. The evaluation tools may not have been reliably developed and validated, and lack of comparability makes it more difficult to draw together the available evidence. The studies were not formally quality assessed therefore caution is advised when considering these findings.

The included studies have been broadly grouped according to the model of follow-up being evaluated.

### **Problem-oriented and informal follow-up**

Risk-based follow-up or problem oriented follow-up appears to be a common strategy. The observational studies identified broadly suggest that patients who are not routinely followed-up may in fact benefit from problem-oriented or informal follow-up programmes.<sup>16,17</sup> Even where patients feel they are not showing late-effects signs there may be relevant complications which will benefit from medical attention.

### **Shared-care model**

The observational study by Blaauwbroek et al. shows a shared care model combining hospital clinic-based with family doctor provision is both feasible and acceptable to the majority of both patients and family doctors.<sup>18</sup> This paper represents an important step in prospective evaluation of service provision, and highlights the need for comparative studies in this area.

### **Multidisciplinary clinic**

The 'CHIP' multidisciplinary clinic model appears to overcome the gap for patients with multiple late-effects where an annual visit or 'traditional late effects clinic' is not sufficient. Carlson et al.'s descriptive paper with audit of patient satisfaction showed families were unanimously satisfied and would use the clinic again. Benefits around scheduling appointments were also identified.<sup>19</sup>

### **Late effects hospital based clinics**

An observational study and two audits around hospital-based late-effects clinics varied in their aims and the services provided. All appeared to offer access to more than one health care professional (usually a specialist nurse plus a physician or consultant), and seem to be a reduced version of the newer multidisciplinary clinic models.<sup>20-22</sup>

### **Paediatric versus adult clinic (hospital based)**

This study compared predictors of patient satisfaction in attendees of a traditional paediatric late-effects clinic and a multi-disciplinary adult setting clinic.<sup>23</sup> Overall survivors were satisfied with the care they were receiving, there was no evidence that either group was more or less informed or felt at-risk to future problems. Survivors who understood the purpose of follow-up care was for clinical support were more satisfied than those expecting psychological support. It was aspects of clinic organisation rather than setting or clinic type which seemed to influence patient satisfaction.

### **Overall findings**

The heterogeneity of the evaluation tools and follow-up schemes, make it difficult to produce an overall description of an effective programme. However it appeared that clinical care was valued highly and supportive care was perceived as more important by patients experiencing more late-effects symptoms and requiring more interventions.

The studies present an apparent contradiction: that some patients who were not followed-up may have been receiving inadequate care; yet there may also be a sub-group of patients for whom long-term follow-up is not an essential part of care. These findings may represent different sub-groups of patients, or they may truly be conflicting results from different follow-up and evaluation programmes.

### **CONCLUSIONS**

Despite a rigorous search, no studies presenting comparative data from retrospective or prospective groups were located. Therefore the conclusion must be that there is too little evidence to firmly answer the research question. However the review has identified areas for focussing future research efforts.

### **IMPLICATIONS FOR RESEARCH**

The nature of the studies presented illustrates the limitations of the research currently available and should provide the impetus for the development of a comprehensive programme of research in this area.

There is a recognition that life-long follow-up is necessary to improve detection of the late-

effects of treatment and to provide information and advice to childhood cancer survivors.<sup>11, 24</sup>

The purpose of long-term follow-up of survivors of childhood cancers has several goals, including: the detection and treatment of late effects; support and advice; and ongoing health education.<sup>25</sup> The long term provision of follow-up for all survivors in health care settings is likely to become unfeasible given increasing numbers of childhood cancer survivors.<sup>20</sup> To address this flexible models of risk stratification have been proposed, for example:

- Level 1 - postal or telephone follow-up following initial treatment involving surgery or low-risk chemotherapy;
- Level 2 - nurse or GP-led follow-up following chemotherapy and/or low-dose radiation; and
- Level 3 - specialist medical follow-up following radiotherapy or megatherapy.<sup>26</sup>

Flexibility within these categories is necessary to take account of the diverse physical and psychological late effects that can occur as well as survivors' expectations about the kind of care they wish to receive.<sup>27</sup> Ongoing evaluation of the experiences of survivors who are stratified and allocated to the relevant follow-up programme will be important to ensure that the needs of patients are being met in a non-clinical setting.

### **RESEARCH PRIORITIES**

Adequately powered, well-conducted, controlled trials of sufficient duration that directly compare the interventions and comparators specified within this review would provide robust evidence on the optimal follow-up or aftercare for childhood cancer survivors. These trials should be conducted in appropriate settings and focus on meaningful outcomes such as patient experience, detection of morbidities and mortality rates. The economic implications of these interventions should also be assessed.

A priority-setting exercise should be conducted to assess which, if any, aspect of health promotion strategies could be subject to systematic review.

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