

# 16TH PANCare MEETING

VIENNA, SEPTEMBER 23 - 25, 2015





## PREFACE

Julio D'Angio: "To cure is not enough".

It was a great pleasure for me to host the 16th PanCare Meeting in Vienna, September 23–25, 2015.

Since the first Meeting in 2008 in Lund, the number of participants was constantly increasing and there were 152 participants in Vienna.

Before becoming an association last year PanCare was built up by a group of people interested in long-term follow-up care and research after cancer in childhood and adolescence. It became obvious that the well-established and well-functioning structures in paediatric oncology are limited concerning the needs of adolescents and adults after cancer in childhood and adolescence. New concepts became also necessary for research of long-term sequelae.

The close collaboration between the different professional disciplines and the intensive exchange with survivor organisations form the unique profile of the PanCare Association. This is traditionally reflected in the diversity of the participants and the programmes of the PanCare meetings.

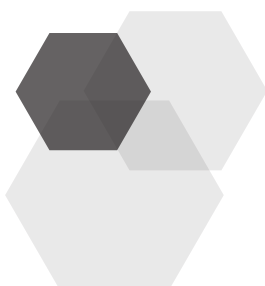
For the meeting in Vienna the focus was on three issues, which are closely related to each other:

- Late effects during adulthood after childhood cancer:
- Transition from the paediatric to the adult health care system
- Concepts for collaborations between paediatricians and physicians in different disciplines in adult medicine

Many thanks to all colleagues who contributed to make this meeting a successful one. All abstracts are published in these proceedings; the chapters represent the topics of the meeting.

The development of research in the field of late effects increased in volume and level of quality in the last years, this was represented in the research papers of our meeting. The collaborations in the PanCare group are one of the reasons for this successful development. The fact that PanCare has two EU-projects running and is collaborating in other EU-projects underlines the high level of research which became possible under the umbrella of the PanCare association.

We will inform all participants of the meeting about the release of the proceedings and the access will be possible on the PanCare website.



Eva Frey, MD  
Local Host  
Chair of the Scientific Committee

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## RESEARCH OF LATE EFFECTS DURING ADULTHOOD AFTER CHILDHOOD CANCER

### L2.3 Clinical questions about late effects, especially genetic predisposition

Thorsten Langer<sup>1</sup>, Oliver Zolk<sup>2</sup>

<sup>1</sup>LESS Center, Children's Hospital, University Clinic Schleswig-Holstein, Campus Lübeck, Germany, <sup>2</sup>Institute of Pharmacology of Natural Products and Clinical Pharmacology, University Hospital Ulm, Germany

### L2.4 How can we facilitate research in daily practice of a late effects service for adult CCS

Diana Greenfield

Weston Park Hospital Sheffield, United Kingdom

### 3.1 Ototoxicity in Swiss childhood cancer long-term survivors

A. Weiss<sup>1</sup>, R. Kuonen<sup>1</sup>, R. Kasteler<sup>1</sup>, L. Wengenroth<sup>1</sup>,  
K. Scheinemann<sup>2</sup>, M. Grotzer<sup>3</sup>, M. Kompis<sup>4</sup>, C. Kuehni<sup>1</sup>

<sup>1</sup>University Bern, Institute of Social and Preventive Medicine, Bern, Switzerland, <sup>2</sup>Division of Pediatric Hematology/Oncology, Children's Hospital Lucern, Lucerne, Switzerland, <sup>3</sup>Department of Pediatric Oncology, University Children's Hospital Zurich, University of Zurich, Zurich, Switzerland, <sup>4</sup>Department of ENT, Head and Neck Surgery, University Hospital Bern, Bern, Switzerland

### 3.2 Respiratory disease after childhood cancer in Switzerland

R. Kasteler<sup>1</sup>, A. Weiss<sup>1</sup>, R. Kuonen<sup>1</sup>, G. Sommer<sup>1</sup>, L. Wengenroth<sup>1</sup>, P. Latzin<sup>2</sup>, N. X. Von der Weid<sup>3</sup>,  
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### 3.3 (In search of potential predictors of) long term impact of chemotherapeutics on the brain of childhood leukemia survivors

I. Elens<sup>1</sup>, J. Lemiere<sup>1</sup>, E. Bossuyt<sup>1</sup>, C. van Soest<sup>1</sup>, H. Vanderstichele<sup>2</sup>, J. Cools<sup>1</sup>, S. W. Van Gool<sup>1</sup>

<sup>1</sup>KU Leuven, Leuven, Belgium, <sup>2</sup>ADx Neuroscience, Ghent, Belgium.

### 3.4 Metabolic risk is associated with reduced hypothalamic volume in Acute Lymphoblastic Leukemia survivors 34 years after cranial radiotherapy

C. Follin<sup>1</sup>, S. Gabery<sup>2</sup>, Å. Petersen<sup>2</sup>, P. Sundgren<sup>3</sup>, I. Björkman-Burtcher<sup>3</sup>, J. Lätt<sup>3</sup>, P. Mannfolk<sup>3</sup>,  
E. Erfurth<sup>4</sup>

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### 3.5 Ultrasound surveillance for radiation-induced thyroid carcinoma in adult survivors of childhood cancer

E. Brignardello<sup>1</sup>, F. Felicetti<sup>1</sup>, A. Castiglione<sup>2</sup>, M. Gallo<sup>3</sup>, F. Maletta<sup>4</sup>, G. Isolato<sup>5</sup>, E. Biasin<sup>6</sup>, F. Fagioli<sup>6</sup>,  
A. Corrias<sup>7</sup>, N. Palestini<sup>8</sup>

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### 3.6 The long-term impact of TBI in pediatric stem cell transplant on the thyroid gland

N. Zubarovskaya, E. Güclü, C. Dampf, U. Poetschger, C. Peters, A. Lawitschka

St. Anna Children's Hospital, Medical University, Vienna

### 3.7 Musculoskeletal sequelae and rehabilitation of childhood solid tumours survivors

A. Petrichenko, E. Bukreeva, N. Ivanova, T. Sharoev, A. Prityko

Child Health Care Research Clinic, Moscow, Russian Federation.

- L3.8 Lecture: Aesthetic and functional impairments – underestimated late effects**  
**Manfred Frey, Igor Pona, Eva Frey**  
 International Center for Facial Nerve Surgery Vienna, Department of Plastic and Reconstructive Surgery, Medical University Vienna, St. Anna Children's Hospital, Medical University Vienna, Austria
- 4.1 Health of offspring of former childhood cancer patients – a survey**  
**A. Borgmann-Staudt, M. Balcerek, E. Korte, R. Schilling**  
 Charité, Berlin, Germany
- 4.2 (8+5') Quality of life for survivors of childhood cancer**  
**S. De Clercq<sup>1</sup>, N. Van Damme<sup>1</sup>, K. Vande loock<sup>1</sup>, K. Emmerechts<sup>1</sup>, E. Van Eycken<sup>1</sup>, Members of the steering committee<sup>2</sup>, BSPHO<sup>3</sup>**  
<sup>1</sup>Belgian Cancer registry, Brussels, Belgium, <sup>2</sup>Childhood Cancer Centers, Brussels/Ghent/Leuven/Antwerp/Liège, Belgium, <sup>3</sup>BSPHO, Brussels, Belgium.
- 4.3 Do childhood cancer survivors participate in physical education at school and club and leisure time sports activities? – An analysis of the current status and remaining barriers**  
**S. V. Kesting<sup>1</sup>, M. Götte<sup>1</sup>, C. C. Seidel<sup>2</sup>, D. Rosenbaum<sup>2</sup>, J. Boos<sup>1</sup>**  
<sup>1</sup>Department of Pediatric Hematology and Oncology, University Hospital, Münster, Germany, <sup>2</sup>Institute for Experimental Musculoskeletal Medicine, Movement Analysis Lab, University Hospital, Münster, Germany.
- 4.4 (8+5') International project on prospective analysis of radiation toxicity in childhood and adolescence (IPPARCA): patients will profit from results**  
**P. Gehoff<sup>1</sup>, H. Christiansen<sup>1</sup>, D. Steinmann<sup>1,2</sup>, H. Eich<sup>3</sup>, K. Susek<sup>3</sup>, H. Wolters<sup>3</sup>, U. Martinsson<sup>4</sup>, H. Magelssen<sup>5</sup>, N. Willich<sup>2</sup>**  
<sup>1</sup>Department of Radiotherapy, Hannover Medical School, Hannover, Germany, <sup>2</sup>Department of Radiotherapy, University Hospital Muenster, Muenster, Germany, <sup>3</sup>University Hospital Muenster, Muenster, Germany, <sup>4</sup>Department of Oncology, University Hospital Uppsala, Uppsala, Sweden, <sup>5</sup>Department of Radiooncology, University Hospital Oslo, Oslo, Norway.
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- TRANSITION – TOOLS AND BENCHMARKS**
- 5.2 Parents' preferences for the organization of long-term follow-up of childhood cancer survivors**  
**J. Vetsch<sup>1</sup>, C. Rueegg<sup>1</sup>, L. Mader<sup>1</sup>, E. Bergstraesser<sup>2</sup>, M. Diezi<sup>3</sup>, C. Kuehni<sup>4</sup>, G. Michel<sup>1,4</sup>**  
<sup>1</sup>University of Lucerne, Lucerne, Switzerland, <sup>2</sup>University Children's Hospital Zurich, Zurich, Switzerland, <sup>3</sup>Paediatric Hemato-Oncology Unit and Division of Clinical Pharmacology, Lausanne, Switzerland, <sup>4</sup>Institute of Social and Preventive Medicine, Berne, Switzerland.
- 5.3 Advocacy masterclass for survivors of paediatric and adolescent cancers**  
**K. Rizvi, D. Tomai, E. Schipor**  
 Little People Association, Cluj-Napoca, Romania
- 5.4 Network oncological specialist advisory service (NOF)**  
**R. Kampschulte<sup>1</sup>, A. Mohr<sup>2</sup>, U. Neuhaus<sup>1</sup>, K. Kreimeike<sup>1</sup>, D. Reinhardt<sup>3</sup>**  
<sup>1</sup>Netzwerk für die Versorgung schwerkranker Kinder und Jugendlicher e.V., 30625 Hannover, Germany, <sup>2</sup>Medizinische Hochschule Hannover, 30625 Hannover, Germany, <sup>3</sup>Universitätsklinikum Essen, Klinik für Kinderheilkunde III, 45147 Essen, Germany.
- L6.1 Lecture: Psychosocial issues as benchmarks for successful transition from paediatric to adult health care service**  
**Gisela Michel**  
 Health Science and Health Policy, University of Lucerne, Lucerne, Switzerland
- 6.2 Screening for cognitive deficit in 8 to 14 year old children with cerebellar tumours using self-report measures of executive and behavioural functioning and health-related quality of life**  
**K. S. Bull<sup>1</sup>, C. Liossi<sup>1</sup>, J. L. Peacock<sup>2,3</sup>, H. Yuen<sup>1</sup>, C. R. Kennedy<sup>1,4</sup>**  
<sup>1</sup>University of Southampton, Southampton, United Kingdom, <sup>2</sup>King's College London, London, United Kingdom, <sup>3</sup>Guy's and St Thomas' NHS Foundation Trust, London, United Kingdom, <sup>4</sup>University Hospital Southampton NHS Foundation Trust, Southampton, United Kingdom.
- 6.3 Neuropsychological methods to facilitate school participation of pediatric brain tumor patients**  
**T. Pletschko<sup>1</sup>, S. Knasmüller<sup>2</sup>, A. Schwarzingner<sup>1</sup>, L. Weiler<sup>1</sup>, U. Leiss<sup>1</sup>**  
<sup>1</sup>Medical University of Vienna, Vienna, Austria, <sup>2</sup>University of Vienna, Vienna, Austria.

- 6.4 **Childhood cancer survivors' perceptions of quality in endocrine care in Sweden**  
A. Pålsson<sup>1</sup>, T. Wiebe<sup>2</sup>, C. Follin<sup>3</sup>  
<sup>1</sup>Department of Endocrinology, Lund, Sweden, <sup>2</sup>Department of paediatric oncology, Lund, Sweden, <sup>3</sup>Institution of Clinical Sciences, Lund, Sweden.
- 6.5 **Factors associated with specific worries of childhood cancer survivors: a cross-sectional survey in Japan**  
Y. Ishida<sup>1,2,3</sup>, T. Higaki<sup>4</sup>, M. Hayashi<sup>3</sup>, F. Inoue<sup>3</sup>, M. Ozawa<sup>2</sup>  
<sup>1</sup>Ehime Prefectural Central Hospital, Matsuyama, Japan, <sup>2</sup>St. Luke's International Hospital, Tokyo, Japan, <sup>3</sup>Heart Link Working Project, Niigata, Japan, <sup>4</sup>Ehime University Graduate School of Medicine, Toon, Japan.
- 6.6 **Childhood cancer awareness education sessions under Haima CZ**  
L. Cingrossova<sup>1,2</sup>, V. Kotková<sup>2</sup>, T. Pasková<sup>2</sup>, Z. Somosová<sup>2</sup>, J. Kruseova<sup>1</sup>  
<sup>1</sup>Department of Pediatric Haematology and Oncology University Hospital Prague Czech Republic, Prague, Czech Republic, <sup>2</sup>Haima CZ, Association of childhood cancer survivors and parents of childhood cancer survivors, Prague, Czech Republic.
- 6.7 **The psychosocial impact of a peer-support residential for young people treated for retinoblastoma**  
E. Hughes, C. Preece, H. Jenkinson  
Birmingham Children's Hospital, Birmingham, United Kingdom.
- 9.1 **Transition for adult survivors of childhood cancer in Austria: ZONE makes it possible !**  
C. Schneider, S. Karner, A. Kienesberger, L. Kager, W. Holter, E. Frey  
Österreichische Kinder-Krebs-Hilfe, St. Anna Children's Hospital, Medical University Vienna, Austria.
- 9.2 **Program of follow-up of long-term survivors of childhood cancer in Slovenia**  
L. Zadavec Zaletel, A. Vodusek, B. Jereb  
Institute of Oncology, Ljubljana, Slovenia
- 9.3 **Transition – A process not an event**  
E. Potter, L. Soanes, J. Passmore, M. Taj;  
The Royal Marsden NHS Foundation Trust, Sutton, United Kingdom.
- 9.5 **Long term follow up consultation after childhood cancer in France (Rhône-Alpes region): the voices of adults and their general practitioners**  
C. Berger<sup>1,2,3</sup>, L. Casagrande<sup>4,5</sup>, C. Faure-Contier<sup>6</sup>, D. Plantaz<sup>7</sup>, F. Isfan<sup>8</sup>, B. Trombert-Paviot<sup>9,5</sup>, I. Guichard<sup>10</sup>, I. Durieu<sup>11</sup>, P. Cathebras<sup>10</sup>  
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- L9.6 **Lecture: Improving the experience of moving to adult care: developement of benchmarks for transition**  
Faith Gibson  
Clinical Professor of Children and Young People's Cancer Care, Centre for Outcomes and Experience Research in Children's Health, Illness and Disability Great Ormond Street Hospital for Children NHS Foundation Trust, and Department of Children's Nursing, London South Bank University, London, United Kingdom

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## POSTER PRESENTATIONS

- P.01 **No evidence for longterm chemotherapy-induced neurotoxicity for pediatric ALL patients: A prospective study**  
C. Sleurs<sup>1,2</sup>, J. Lemiere<sup>1</sup>, T. Vercruysse<sup>1</sup>, N. Nolf<sup>3</sup>, B. Van Calster<sup>2</sup>, S. Deprez<sup>4</sup>, M. Renard<sup>1</sup>, E. Vandecruys<sup>3</sup>, Y. Benoit<sup>3</sup>, A. Uytendaele<sup>1,2</sup>  
<sup>1</sup>Department of Pediatric Haematology and Oncology, University Hospitals Leuven, Leuven, Belgium, <sup>2</sup>Department of Development and Regeneration, KU Leuven, Leuven, Belgium, <sup>3</sup>Department of Pediatric Haematology and Oncology, Ghent University

Hospital, Ghent, Belgium, <sup>4</sup>Department of Radiology, KU Leuven, Leuven, Belgium.

P.02                    **Functional and clinical long-term outcome of Ewing sarcoma treatment**  
**A. Ranft**, C. Hoffmann, C. Seidel, H. Jürgens, D. Rosenbaum, U. Dirksen

University Hospital, Muenster, Germany.

P.05                    **FeCt – Multicentre Offspring Study – the results of the participating institution St. Anna**  
**Children's Hospital**

**E. Frey**, M. Kalemba-Holzgethan

St. Anna Children's Hospital, Medical University, Vienna



Childhood cancer survival rates are nearly 80% in more developed European countries because of improved therapies and better supportive care. However, the antitumor efficacy of optimized therapy protocols comes at the cost of late adverse effect of treatments, which affects a large proportion of pediatric patients and can have debilitating effects on patients' quality of life. Prominent examples are anthracycline-induced cardiotoxicity, chemotherapy-associated infertility, ifosfamide-related nephrotoxicity or cisplatin-induced ototoxicity. Although clinical risk factors for these complications have been identified, these risk factors alone are insufficient to accurately stratify patients into groups at high and low risk, which may have major implications for patient care. Moreover, the pathophysiology of most of these late complications is not fully understood, which complicates the identification of new risk markers.

Genetic studies may help to identify markers predictive of late adverse effects or could help to elucidate more precise mechanisms. Several candidate gene studies have been conducted with promising results and the first genome-wide association studies are currently performed. For example, recent results demonstrated a highly significant association of single nucleotide polymorphisms (SNPs) in the SLC28A3 gene with anthracycline-induced cardiotoxicity in children (odds ratio, 0.35;  $P = 1.8 \times 10^{-5}$ ) (Visscher et al., 2013)

Validation of initial association findings is essential, as demonstrated by the cisplatin example (Langer et al., 2013). SNPs in the thiopurine methyltransferase (TPMT) gene were significantly linked with the risk of cisplatin-induced ototoxicity. This association was surprising because a mechanistic rationale for the involvement of the enzyme in the cisplatin pathway was missing. On the basis of a single study and with absolutely no mechanistic justification, the US Food and Drug Administration added a warning to the label for cisplatin which linked TPMT genetic variation to ototoxicity.

Subsequently, however, contradictory clinical as well as mechanistic data were presented refuting the association of TPMT polymorphisms with ototoxicity.

Only a small fraction of the total genetic risk for late complications can be explained by the mutations investigated so far in candidate gene association studies. Results from larger, better controlled genome-wide studies will help to identify relevant genetic loci. Toxicogenetic information in the drug label is useful clinically only on the basis of positive replications and with a clear mechanistic justification. It is most likely that patients' genetic information will be used in combination with clinical risk factors to effectively stratify the patient's individual risk for late complications after cancer therapy.

#### References:

Visscher H, Ross CJ, Rassekh SR, Sandor GS, Caron HN, van Dalen EC, Kremer LC, van der Pal HJ, Rogers PC, Rieder MJ, Carleton BC, Hayden MR; CPNDS Consortium (2013) Validation of variants in SLC28A3 and UGT1A6 as genetic markers predictive of anthracycline-induced cardiotoxicity in children. *Pediatr Blood Cancer* 60:1375–81

Langer T, Am Zehnhoff-Dinnesen A, Radtke S, Meitert J, Zolk O (2013)

Understanding platinum-induced ototoxicity. *Trends Pharmacol Sci.* 2013 Jun 13. pii: S0165-6147(13)00094-1. doi:10.1016/j.tips.2013.05.006. [Epub ahead of print]

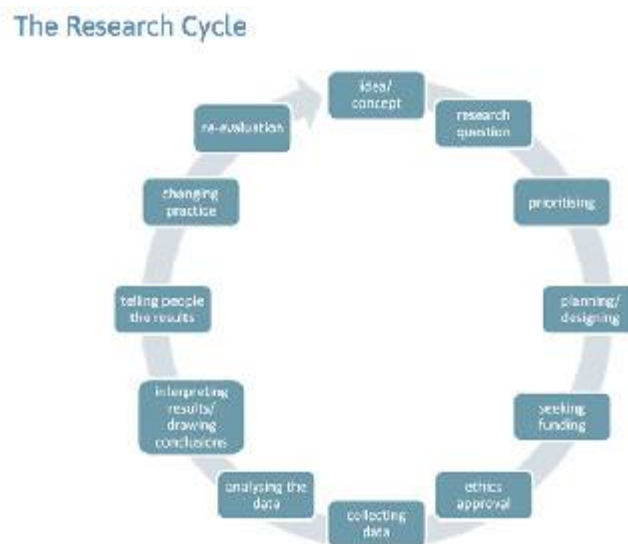
Answer: not alone! This presentation briefly considered the multitude of elements which may facilitate research in daily clinical practice in a late effects clinic, including the obligatory operational components and the crucial individual characteristics, which together will contribute to success.

Firstly and most importantly, a dedicated and committed multi-disciplinary team is a must, comprising both clinical and non-clinical staff representing different levels of seniority, and ideally lead by a clinical academic. The host healthcare institution will preferably already foster an ethos of clinical research by providing a supportive institutional framework for research governance and ethical approval procedures.

Research starts with a research question, often generated from a clinical conundrum, such as an observation in clinical practice. Late Effects Clinic provides us with an endless opportunity for investigating observed clinical conundrums.

Patient and Public Involvement (PPI) is recommended from the outset, particularly one which fosters “co-design”, that is including PPI in all stages as equal contributors, with any previously paternalistic approaches designated to history! Patient awareness of research in the clinical setting can be encouraged as norm. This can be done by promoting research participation in patient information leaflets distributed in advance of late effects clinic and reiterated at clinic attendance.

Knowledge and experience of each stage of the research cycle is a given (see below), with the participating team scheduled dedicated time, individually and collectively, to every stage of the research cycle. This is often the most difficult challenge, with clinical responsibilities understandably taking priority.



(Downloaded from <http://www.pencru.org/research/whatdowemeanbyresearch/>)

Securing adequate research funding is often easier said than done, with identification of possible funding streams the first challenge, and the development and submission of a robust application the next.

But securing funding is only the start. Successful research lies in the preparation with this often requiring as much investment in time than the actual fieldwork (or “doing”). So even before the research comes to the late effects clinic, much of the work has already been done.

Analysing data needs expert involvement, away from the clinical setting but interpretation of results and drawing conclusion a collective effort. Writing for publication is hugely skilled and once again, this part of the research cycle may be as time-consuming as the “doing”. “A third: A third: A third” is a good guide (preparation: doing: dissemination).

Despite the time lag from completing the field work, when it comes to dissemination of findings, it's good practice to share with both participating patients and staff from the clinical setting. This will promote a sense of ownership and engagement and may help to keep staff up to date and motivated to change. And the latter is the next step.

Changing practice to ensure research outcomes are adopted and integrated into clinical practice is easier if the entire team is engaged, motivated and valued. Changing practice is not easy in a busy pressured clinical service. Re-evaluation and patient feedback are also important to ensure effective implementation.

Ultimately, undertaking research in clinical practice requires strong leadership, skill, a combination of clinical and academic expertise, a committed team, partnership with patients, conviction, perseverance and much elbow grease!



**Introduction:** Childhood cancer treatment, particularly with platinum compounds, can be toxic to the ear. While hearing impairment shortly after treatment has been extensively investigated, there are little data on long-term outcomes. Therefore, our aims were: 1) to determine prevalence of hearing loss and tinnitus in long-term survivors of childhood cancer, compared to their siblings; and 2) to describe the associations with cancer treatment.

**Methods:** As part of the Swiss Childhood Cancer Survivor Study, we sent a detailed questionnaire to all 5-year survivors of childhood cancer, diagnosed 1976–2005, who were registered in the Swiss Childhood Cancer Registry. We compared prevalence of reported hearing loss and tinnitus between survivors and siblings and analyzed associations with treatments (platinum compound, radiation to inner ear) by using multivariable logistic regression.

**Results:** 2380 survivors (response rate 71%) and 864 siblings (56%) returned the questionnaire. Mean (SD) age at survey for survivors was 24 (9) years and mean time since diagnosis 16 (7) years. All diagnostic groups were included: the largest being leukemia (32%), lymphoma (18%), and CNS tumors (15%). Survivors reported significantly more often hearing loss than siblings (10.0% (95% CI 8.8–11.3) vs. 3.3% (2.2–4.8),  $p < 0.001$ ). Prevalence of tinnitus did not differ (4.5% vs. 4.5%,  $p = 0.955$ ). Among the 229 survivors with hearing loss, 125 reported the date of onset of hearing loss: in 44% hearing loss appeared after 5 or more years post-diagnosis. In a multivariable logistic regression adjusted for age at treatment and gender, children exposed to radiation to the inner ear (OR 1.48), platinum compounds (OR 5.76) or both (OR 14.70) were at increased risk for hearing loss ( $p < 0.001$ ). Radiation to the inner ear (OR 1.45), treatment with platinum compounds (OR 1.32), or a combination of both (OR 3.13) was associated with tinnitus ( $p = 0.042$ ).

**Conclusion:** This study provides first data on ototoxicity after childhood cancer in Switzerland. It shows, in accordance with international studies, the relevance of ototoxic late effects: 1) Survivors reported more often hearing loss than siblings and 2) radiation to inner ear and treatment with platinum compound were associated with hearing loss. In a considerable proportion of survivors (44%) the onset of hearing loss was 5 or more years after diagnosis. The findings indicate a long-term ototoxic effect of these treatments and thus the need of a long-term monitoring system in patients at risk.

**Background:** Respiratory dysfunction is a severe late complication of childhood cancer treatment, with survivors having a tenfold increased risk for a late pulmonary death. Little data are available on early respiratory disease. Therefore, we aimed 1) to compare prevalence of early respiratory diseases between long-term survivors and their siblings; and 2) to determine risk factors for respiratory disease.

**Methods:** As part of the Swiss Childhood Cancer Survivor Study, we sent a detailed questionnaire to all 5-year survivors of childhood cancer, diagnosed 1976–2005, registered in the Swiss Childhood Cancer Registry. We compared prevalence of reported respiratory diseases (asthma, hay fever, pneumonia, sinusitis, chest wall abnormalities, and pulmonary fibrosis) between survivors and siblings. We defined two risk groups for respiratory disease: i) survivors treated with methotrexate, alkylating agents and/or bleomycin treatment and ii) survivors with irradiation to the chest. We determined risk factors for respiratory diseases using multivariable logistic regression.

**Results:** 2380 survivors (response rate 71%) and 864 siblings (53%) returned the questionnaire. Mean age at survey was 24±9 (SD) years. Survivors generally reported more respiratory diseases than siblings (40% vs. 35%, p-value=0.016). They reported significantly more recurrent pneumonias (9% vs. 6%, p=0.004), recurrent sinusitis (9% vs. 7%, p=0.009) and chest wall abnormalities (2% vs. 0.3%, p<0.001). Hay fever, asthma and fibrosis were equally reported. 1368 survivors (58%) had received pulmotoxic treatment (i and/or ii), 1246 (52%) pulmotoxic chemotherapy (i), 299 (13%) irradiation to the chest (ii), 177 (7%) a combination of both (i+ii). Chest wall abnormalities were associated with irradiation to the chest (OR 3.80, p<0.001), after adjusting for age at treatment and sex.

**Conclusion:** This is the first study to describe respiratory disease after childhood cancer in Switzerland. More than half of survivors had received potentially pulmotoxic treatment. This and the higher prevalence of recurrent pneumonia and sinusitis, chest wall abnormalities and overall respiratory diseases shows the need for clinical and objective validation with lung function measurements and more detailed risk stratification including cumulative doses of irradiation and chemotherapeutic drugs.

**Background:** Contemporary treatment can cure children with NHL or ALL. However, chemotherapy-induced neurotoxicity – a condition in adult medicine termed ‘chemobrain’ – causes long-lasting functional deficits in some survivors. Besides primary cure, early identification and subsequent care for these individuals must therefore be focus of research.

Previously, we reported increased levels of biomarkers of neurotoxicity in the cerebrospinal fluid (CSF) during treatment and short-term information processing difficulties in survivors.

The methylenetetrahydrofolate reductase (MTHFR) enzyme plays a key role in the folate metabolism. Individuals with polymorphisms causing lower activity of this enzyme have a higher risk for methotrexate-induced neurotoxicity.

#### Aims:

1/ To describe long-term neurocognitive functioning of childhood ALL and NHL survivors.

2/ To identify predictors of neurocognitive outcome.

**Methods:** CSF-Tau and phospho-Tau were chronologically measured during treatment (1996–2004) in respectively 28 and 7 patients with ALL and NHL. The reaction time (RT) on the ANT (Amsterdam Neuropsychological Task) and questionnaires was compared with sex- and age-matched controls, and in survivors, the genotype on position 677 and 1298 of the MTHFR-gene was analyzed.

#### Results:

1/ Description of long-term neurocognitive functioning of childhood ALL and NHL survivors

Patients performed equal to controls concerning baseline RT and number of errors. However, they were significantly slower on tasks assessing focused ( $p = 0.001$ ) and divided ( $p = 0.049$ ) attention as well as cognitive flexibility ( $p < 0.001$ ), suggesting white-matter dysfunction. These observations were confirmed by higher scores on subscales of self-reported cognitive dysfunctions, whereas there were no significant differences on self-reported overall quality of life, or depressive or anxious state.

2/ Exploration of predictors of neurocognitive outcome

Treatment related factors (ALL vs NHL, risk classification, protocol, randomization to dexamethasone or prednisolone) could not predict the RT.

Survivors diagnosed before the age of five tended ( $p = 0.15$ ) to perform slower.

The MTHFR1298 CC genotype was linked with higher levels of neurotoxicity markers during the induction phase of the treatment as compared to the heterozygote (AC) and wild-type (AA) variant ( $p = 0.024$  and  $0.020$  respectively), possibly reflecting higher vulnerability to MTX of this genotype.

#### Conclusions

We confirm the long-term impact of chemotherapeutics on the developing brain. Since treatment related factors alone could not predict the RT, this study emphasizes the need to identify new markers for chemotherapy-induced neurotoxicity. We therefore explore a potential link between the neurotoxic hit during chemotherapy, the subsequent biomarker profile, the folate metabolism and the final neurocognitive outcome.

Identification of polymorphisms or biomarker profiles during chemotherapeutic treatment predictive for neurocognitive dysfunctions might enable us to early support susceptible children with appropriate learning programs. Validated biomarkers might become key parameters to evaluate – apart from efficacy – the toxicity of new chemotherapeutics.

### Metabolic risk is associated with reduced hypothalamic volume in Acute Lymphoblastic Leukemia survivors 34 years after cranial radiotherapy

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**Background:** Cancer survivors exposed to cranial radiotherapy (CRT) are at particularly high risk and survivors of childhood brain tumors with hypothalamic damage, either due to tumor, surgery or CRT, are at increased risk for obesity. It is established that the acute lymphoblastic leukemia (ALL) survivors, treated with CRT have GH-deficiency (GHD), obesity, lipid abnormalities and insulin resistance. Further, leptin resistance has been recorded among GH deficient ALL survivors and in particular, among ALL women, indicating a radiation-induced hypothalamic dysfunction. The hypothalamus (HT) is a complex brain region involved in endocrine function and metabolic control. The HT might be affected and behind the persistent metabolic problems after CRT in ALL survivors. Further, we anticipated that peptides like insulin, leptin and ghrelin might be affected and possibly more among ALL women compared to ALL men. Thus, our aim was to assess the volume of the HT in relation to metabolic parameters including insulin, leptin and ghrelin in ALL survivors.

**Method:** Thirty-eight ALL survivors (21 women), median-age of 38 (27–46) years were investigated 34 years after ALL diagnosis. All had been treated with a CRT dose of 24 Gy and were on complete hormone supplementation. Comparison was made with 31 healthy matched controls. Assessments of BMI (kg/m<sup>2</sup>), waist (cm), fat mass (DXA/kg), fat free mass (kg), plasma (p)-glucose (mmol/L), p-insulin (mIE/L), Homa-Index, serum (s)-leptin (µg/L) and s-ghrelin (ng/L) was performed. Magnetic resonance imaging (MRI) was performed to conduct volumetric analysis of the HT.

**Results:** S-leptin/kg fat mass levels ( $r=-0.4$ ,  $P=0.04$ ) and fat mass ( $r=-0.4$ ,  $P=0.01$ ) were negatively correlated with the HT volume among the 34 ALL survivors (figure 3), but not among the controls ( $P>0.3$ ). There was a trend of a smaller HT volume among the ALL women compared to gender matched controls (846 vs 869 mm<sup>3</sup>,  $P=0.06$ ). There was a significantly negative correlation of serum ghrelin to age at diagnosis ( $r=-0.4$ ,  $P=0.03$ ) and a positive correlation of serum ghrelin to follow-up time after diagnosis ( $r=0.4$ ,  $P=0.03$ ) (figure 4). Among female ALL survivors, but not among ALL men, compared to controls (all  $P<0.01$ ), significantly higher BMI (27.9 vs 22.6 kg/m<sup>2</sup>), waist (89 vs 79 cm), fat mass (29.9 vs 22.4 kg), p-insulin (10 vs 6 mIE/L), Homa-Index (0.15 vs 0.07), leptin/kg fat mass (1.09 vs 0.6) and s-ghrelin (1560 vs 993 ng/L) and significantly lower fat free mass (35.4 vs 41.6 kg) were recorded.

**Conclusions:** 34 years after ALL diagnosis Leptin/kg fat mass and fat mass was associated with a reduction in HT volume. ALL women treated with CRT are at high risk of metabolic abnormalities. S-ghrelin levels increased with time since diagnosis and with low age at diagnosis, underscoring the possibility of a hypothalamic dysfunction behind the metabolic problems.

## Ultrasound surveillance for radiation-induced thyroid carcinoma in adult survivors of childhood cancer

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**Introduction:** Childhood cancer survivors (CCS) treated with radiotherapy to the head, neck or upper thorax are at increased risk of developing a thyroid cancer as second neoplasm, but the optimal surveillance strategy for these subjects is still debated. In our clinical practice, beside neck palpation we routinely perform thyroid ultrasound (US) to all patients at risk. Here we describe the results obtained using this approach.

**Patients and Methods:** We considered all CCS referred to our long term follow-up unit from November 2001 to September 2014. Thyroid US, which is always performed by a specialist with high expertise in the specific field of cancer survivors, usually starts 5 years after radiotherapy and is repeated every three years, if negative. Thyroid nodules are considered when solid at US and with major diameter  $\geq 0.5$  cm. If a nodule is found, thyroid US is repeated every 12 months (or less) or fine-needle aspiration (FNA) is performed on nodules bearing a combination of US characteristics suggestive for malignancy.

**Results:** Among 197 CCS previously irradiated to the thyroid gland, 74 patients (37.5%) developed thyroid nodules, and FNA was performed in 35 of them. In 11 patients the cytological examination was suspicious (TIR 4, n=4) or diagnostic (TIR 5, n=7) for malignancy, whereas a follicular lesion (TIR 3) was diagnosed in other 9 patients (Table 1). Patients with TIR 4/5 cytology were operated and in all cases thyroid cancer diagnosis was confirmed by histological examination. The 9 patients with TIR 3 cytology also underwent surgery and a papillary thyroid carcinoma was diagnosed in 3 of them. The prevalence of thyroid cancer was 7.1%, with a male / female ratio of 9/5. The median elapsed time between radiotherapy and the thyroid cancer diagnosis was 13.08 years (range 8.22 – 23.65 yrs). Tumor size ranged between 4–25 mm, but 6 cancer (43%) were classified T3 because of extra-thyroidal extension. Six patients had nodal metastases at diagnosis; in 8 patients (57%) the tumor was multifocal (Table 2). At the time of the study all patients are disease free, without any evidence of surgery complications.

**Conclusions:** We found a high prevalence of radiation-induced thyroid cancer. Histological features of these tumors (capsule infiltration, multifocality and nodal metastases at diagnosis) indicates that most of them, even if early-diagnosed, are not indolent cancers. Therefore, if performed by experienced clinicians (to avoid unnecessary procedures and decrease psychological stress), we think that US surveillance may be suitable in CCS previously treated with radiotherapy to the head, neck or upper thorax.

Table 1. Clinical characteristics

	N. (Total=197)	%
Nodules		
no	123	62.44
yes	74	37.56
FNA		
no	162	82.24
yes	35	17.76
TIR 2	15	
TIR 3	9	
TIR 4	4	
TIR 5	7	

Table 2. Patients with thyroid cancer.

	Sex	Pediatric Cancer	Age at pediatric cancer	Radiotherapy (Gy)	Thyroid Cytology	Thyroid Histology	Variants	Size (mm)	Multifocality	Vascular Invasion	Extra-thyroidal extension	T	N	M	Age at thyroid cancer (yrs)	Elapsed time (yrs)
1	M	Hodgkin Lymphoma	13.7	Mantle (20)	TIR 5	Papillary Thyroid Cancer (PTC)	Classical	16	yes	yes	yes	3	1b	0	26.1	13.1
2	F	Acute lymphoblastic leukemia	1.3	Cranial (18)	TIR 3	PTC	Classical	13+5	yes	no	no	1b	0	0	21.7	21
3	M	Hodgkin Lymphoma	11.4	Mantle (20)	TIR 5	PTC	Classical	9	no	no	no	1a	0	0	19.5	8.6
4	M	Acute lymphoblastic leukemia	10.1	Cranial (18)	TIR 4	PTC	Classical	13	no	no	no	1b	0	0	22.3	12.9
5	M	Neuroblastoma	3.5	TBI (14)	TIR 4	PTC	Classical+solid	23	yes	yes	yes	3	1b	1	21.2	18.3
6	F	Acute lymphoblastic leukemia	8.8	Cranial (18) + TBI (14)	TIR 3	PTC	Classical + follicular	19 + 3 + 2	yes	no	yes	3	X	X	37.7	29.9
7	M	Acute lymphoblastic leukemia	12.6	TBI (12)	TIR 4	PTC	Classical	7 + 6	yes	yes	yes	3	1b	0	22.0	10.0
8	M	Acute lymphoblastic leukemia	3.4	Cranial (18) + TBI (12)	TIR 5	PTC + Follicular Thyroid Carcinoma (FTC)	Classical	25 (PTC) + 4 (FTC)	yes (PTC) no (FTC)	yes	no	1a (PTC) 2 (FTC)	0	0	17.2	14.3
9	F	Hodgkin Lymphoma	17.3	Cervical (21.6)	TIR 5	PTC	Classical	9	no	no	no	1a	0	0	24.9	8.2
10	M	Acute lymphoblastic leukemia	2.4	TBI (12)	TIR 4	PTC	Follicular	7+3+8+5+2	yes	yes	no	1a	1a	0	16.9	14.9
11	F	Hodgkin Lymphoma	18	Mediastinal (40)	TIR 5	PTC	Sclerosing	13	no	yes	yes	3	1a	0	40.5	23.6
12	F	Acute lymphoblastic leukemia	7.5	TBI (12)	TIR 5	PTC	Classical	25 + 21	yes	yes	yes	3	1b	0	20.0	13.1
13	M	Acute lymphoblastic leukemia	3.2	TBI (12)	TIR 3	PTC	Classical	4	no	no	no	1a	0	X	23.5	20.9
14	M	Ependymoma	4.7	Cranial (59.4)	TIR 5	PTC	Follicular	8	no	no	no	1a	X	X	13.5	9.2

Advances in techniques, supportive care and increasing indications have resulted in a growing number of long-term survivors after paediatric allogeneic stem cell transplantation (HSCT) shifting aftercare and late effects (LE) into the focus of clinical research. Thyroid dysfunction (TD) is most common endocrine LE after HSCT especially after conditioning with total body irradiation (TBI), busulfan and cyclophosphamide. We conducted a retrospective study on TD after paediatric HSCT between 1984–2010 following TBI– conditioning. 116 patients (pts) with a median age at HSCT of 12yrs (2–25,9yrs) and median follow-up of 5,6 yrs (3mo–20yrs) could be included. The median age at transplantation was 12yrs (2–25,9 yrs). A total of 109 pts received HSCT for a haematological malignancy and 7 pts for solid tumors. The majority received bone marrow in 56 pts from matched unrelated donor (MUD), in 52 pts from matched sibling donor and 8 autologous HSCT. TBI was given in 57 pts with 9–13Gy in 6 fractions, in 55 pts with 12Gy in 7–8 fractions and 4 pts received 10Gy as a single dose. The cumulative incidence of TD was 53%. The most common disorders after SCT were subclinical hypothyroidism (43%), euthyroid sicca syndrome (33%), followed by overt hypothyroidism (13%), secondary hypothyroidism (6%) and autoimmune thyroiditis. Ultrasound of the thyroid gland was performed in 50 pts with abnormalities in 66%. Of these cases another 66% correlated with any kind of TD. Risk factors for TD were HSCT from MUD, the years of HSCT, TBI performed in 6 fractions, any kind of graft versus host disease (GVHD, both acute and chronic) and acute GVHD grade IV. Age at HSCT was a risk factor for both overt hypothyroidism and euthyroid sicca syndrome. Additive irradiation close to the thyroid gland did not increase the incidence of TD. Most of our findings are in line with published data and underline the importance of long-term follow-up of the thyroid gland during HSCT aftercare but the correlation with GVHD in this context needs further evaluation.

**Introduction:** advances in diagnosis and treatment of childhood cancer have dramatically increased long-term survival and it is now evident that the disease and its treatment can significantly impair long-term health. Childhood solid tumours survivors are known to be at risk for the serious musculoskeletal late effects that may result in disability. In this connection, the purpose of the research is recommendations for screening, prevention, and management of survivors and the evaluation of efficiency of cancer rehabilitation of children treated with intensive chemotherapy, surgery and radiation therapy.

**Material and methods:** 89 children and adolescents with solid tumours were treated between 1987 and 2014 years. The mean age at the date of orthopedic diagnosis was 13,7 years (from 2 till 24 years). Common sites of primary disease include the thorax, abdomen, trunk and extremities. Most often the affected area was the area of the lower extremity – 46,1 % of the cases. According to the histological variation of the tumor 35 % of the patients had morphologically confirmed diagnoses of Ewing sarcoma, 26% – osteosarcoma, 39% – other solid tumours. 25,8% patients had distant metastases. Among them, 56,5% patients had distant metastases, 43,5% patients had multiple metastases. Treatment consisted of neoadjuvant chemotherapy, the radiotherapy of the initial tumor and metastasis left after the induction and/or oncologic surgery and adjuvant chemotherapy. The local control of the tumor consisting of the surgical ablation of the primary lesion and metastases, if the technical opportunity of this stage is available, including limb-sparing procedures. The most common late effects we had observed were: scoliosis – in 83,1% cases, muscular hypoplasia – 74,2%, osteopenia – 52,8%, limb-length discrepancy in spite of usage of growing endoprosthesis – 49,4%, poor joint movement – 41,6%, musculoskeletal deformity – 38,2%. 70,8% patients had from 3 till 8 late effects. We used the NCI Common Terminology Criteria for Adverse Events for reporting. We have not observed serious Adverse Events, like Grade 4: life-threatening consequences and Grade 5: death related to Adverse Events. Among the 20 children who developed the largest number of late effects, are children who received CT and radiotherapy – 10 %, in the group with major surgery – 55%, CT, surgery and radiotherapy – 35 %. This indicate, that some researchers reevaluate the role of radiation therapy in the development of musculoskeletal sequelae. An individual rehabilitation program should include, combined with early mobilization, physical exercise, kinesiotherapy, aquatic rehabilitation and orthopaedic correction, laser therapy, massage, gait training.

**Results:** currently all patients are alive without disease, following up of 7 to 341 months. Long-term survival is possible, even for patients with metastatic disease.

**Conclusion:** interdisciplinary team, provide services that enable patients to achieve their highest functional status to permit them to return to their role in society. We suggest that the planning of an individual rehabilitation program can dramatically increase the quality of life.



**Introduction:** Aesthetic and functional impairments are typical consequences of local treatment as surgical tumour resection or radiotherapy. Therefore these are common late sequelae especially in those tumour types, which need both treatment modalities, surgery and radiotherapy.

Surgery causes a local defect by the tumour resection, a defect in the contouring of the affected area in the face, in the trunk area, or in the extremities. It can also cause a functional deficit, if important motor and sensory nerves are in the resected area or muscles, bones and joints have to be partly or completely resected.

Radiotherapy affects mainly the growth of the radiated area and the quality of skin. Therefore the age at irradiation is important. For children irradiated at younger age the growth retardation of the irradiated area can cause a major aesthetic problem. The best known example is the rhabdomyosarcoma of the orbital region or the growth retardation of the female breast during puberty.

The most challenging areas for aesthetic-reconstructive surgery are the facial area and the female breast, for functional reconstruction it is the facial nerve and reconstruction of motor function and sensibility in the extremities.

**Patients and Methods:** Since 1996 aesthetic and functional impairments caused by paediatric oncological treatment have been addressed by an interdisciplinary approach between the Oncological Department of the St. Anna Children's Hospital, Vienna and the Section of Plastic and Reconstructive Surgery at the Medical School of the Medical University of Vienna. All treated children have been evaluated together already before tumour resection critical for aesthetic or functional impairments to develop a primary treatment plan preventing or reducing these impairments. Secondary reconstruction has been indicated for those children, who developed their aesthetic or / and functional impairments during growth, or primary reconstruction had not been possible, or had not been considered. The arising indication has been monitored at the late effects clinic over the years or was evaluated at the end of puberty and growth at latest.

**Results:** Aesthetic and functional impairments showed to be rather specific for the tumour entities and their standardized oncologic treatment. Reconstructive requirements and possibilities differed significantly between the individual patients. Radiotherapy in addition to surgery showed to be an important negative factor. The severity of aesthetic and functional late effects depended on the age at onset and the local dosage of irradiation. When primary reconstruction has been considered, the decision for treatment recommendation was made within the interdisciplinary tumour board. Even when the results of reconstruction were minor after irradiation compared to patients without, the aesthetic and functional improvements were of great importance and an important help for daily life of the survivors. Often life was drastically changing after reconstruction and psychosocial integration was becoming possible. An individualized plan for reconstruction showed to be essential to meet the complex problems and needs of the individual patient.

**Conclusion:** Based on a twenty-years of increased multidisciplinary attention to aesthetic and functional impairments as late effects of oncological treatment we conclude the important points for a successful prevention and a competent offer of reconstructive surgery:

1. Knowledge of typical late effects
2. Prevention or reduction by primary reconstruction
3. Multidisciplinary approach including plastic and reconstructive surgery for planning tumour resection
4. Long-term follow-up
5. Conclusive evaluation and initiation of reconstruction at the end of paediatric after care
6. Individualized conception of reconstruction

Tumour type	Location	Treatment modalities	Aesthetic impairments	Functional impairments
Rhabdomyosarcoma	Face / neck	Surgery RXT (EBRT, Brachytherapy)	Facial palsy, lid ptosis asymmetry, deformities, radioderm, maxillary dysfunction	
	Thorax	Surgery / RXT	Asymmetry, deformities, radioderm	
	Extremities	Surgery, RXT		Motor function Sensibility
Ewingsarcoma	Thorax	Surgery/RXT	Asymmetry, Deformities, radioderm	
	Extremities	Surgery/RXT		Motor function sensibility
Osteosarcoma		Surgery	Asymmetry, deformities	Motor function sensibility
Brain tumours: Medulloblastoma		Surgery/RXT	Radioderm, asymmetry, deformities	Facial palsy, Neurological deficits



**Background:** With rising survival rates, quality of life after treatment of former childhood cancer patients has gained increasing interest. Being able to fulfill own family planning and the health of own offspring is of major relevance to survivors. Previous studies revealed significant numbers of survivors fearing possible health impairment in their children. Overall, studies regarding offspring of former patients have not shown an increased risk for genetic instability, malformations or non-hereditary cancer (Nagarajan 2005). In 2010/2011 we conducted a pilot survey on the general health status of former childhood cancer patients' offspring in Berlin. Since 2013 this survey is being conducted as a multicenter offspring study with participating centers in Germany, Austria, Czech Republic, Poland and Switzerland. The study examines if there are differences regarding diseases, well-being, health care utilization and health-related behavior between former patients' and siblings' offspring and children of the general population. In the following, methods and patient characteristics of the first wave of our nationwide survey in Germany are presented.

**Methods:** Former patients in Germany that were known to have a child of their own due to previous studies on fertility (Balcerek 2012) received a 10-paged questionnaire on their offsprings' health via the German Childhood Cancer Registry after approval of the ethical committee of the Charité–Universitätsmedizin Berlin. Questionnaires were strongly based on the KiGGS study on children's health in the German general population of the Robert–Koch Institute (2003–2006, n=17.641) for data comparability. Data analysis was conducted with the statistical package SPSS Version 22. A non-responder analysis was conducted regarding sex, age at diagnosis and time point of the survey, diagnosis and graduation.

**Results:** 393 former patients known to have a child of their own were applicable for participation. 254 (65%) of these patients participated in the first wave of our offspring study in Germany. In total, questionnaires for 418 children were answered. Participants were more likely to be female (185/254, 73%). The median age of participants at diagnosis was 10 years, at the time point of survey 31 years. The median age of the offspring was seven years (range 0–21 years). 48/254 participants (19%) achieved a secondary modern or intermediate school-leaving certificate (107/254, 42%), and 98/254 (39%) High School Diploma.

A non-responder analysis showed no differences according to age at diagnosis and survey. Differences were found in sex ( $p<0.01$ ), graduation ( $p<0.05$ ) and diagnosis of soft tissue tumor ( $p<0.05$ ) with female, higher educated and patients diagnosed with soft tissue tumor being more likely to respond.

**Conclusion:** The health of offspring is a topic of major relevance to former childhood cancer patients. Further information on the general health status is currently being collected in a collective of estimated 1.500 offspring of former childhood cancer patients in Europe with first results being expected starting in 2016

Although childhood cancer is relatively rare and five-year survival attains 84% or more, childhood cancer survivors are subject to increased morbidity and mortality, compared to the general population. Up to 90% of survivors suffer some late effect at one point in their lives, due to the treatment they received. These late effects (or long-term side effects) can occur months or years after treatment has ended and often pose a substantial burden on survivors, negatively impacting their quality of life.

Although most of the (potential) late effects are already known, sparse information is available concerning their exact prevalence on a national basis. The Belgian Cancer Registry receives information on all (adult and pediatric) cancers since 2004 (for the entire country), and is now planning to collect the data concerning acute and late effects that have been encountered by childhood cancer survivors (age at diagnosis: 0–16 years old) in the months respectively years following their cancer treatment.

These effects will be registered by the treating physician through an online registration system. In the first year after the end of treatment, the following items will be registered: pathology data of the tumor, the presence of congenital and pre-existing diseases or conditions, information concerning the received treatment(s) and the acute effects that have been reported by the patient during and in the first months following treatment.

Besides this initial registration, 'follow-up' registrations will be completed by the physician, reporting the late effects that have emerged since the last notification of effects. The registration of late effects will be repeated every 5 years. Follow-up clinics will be installed, responsible for the organization and coordination of follow-up visits to which the cancer survivors will be invited.

Such a national registration of late effects in the cohort of childhood cancer survivors will provide more complete and unbiased information of the side effects, suffered by survivors following cancer treatment. Furthermore, 'new' late effects might emerge as well as unknown associations between specific congenital or pre-existing conditions and the incidence of certain cancer types or late effects can be established. Additionally, the differential occurrence of late effects, evoked by different therapies, provides a useful tool to evaluate and compare such therapies, in terms of their side effects. This might allow the preference of one therapy over others for certain diagnoses or distinct patient groups, as well as aid in the development of new or personalized therapies with a lesser impact on the quality of life of survivors.

The results, obtained for the Belgian population of childhood cancer survivors, will also be compared with those from other (international) institutes and initiatives. International collaborations can thus be established, providing a larger number of survivors (and hence: data) for inclusion in scientific studies, rendering more accurate outcomes and analyses.

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### Do childhood cancer survivors participate in physical education at school and club and leisure time sports activities? – An analysis of the current status and remaining barriers

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**Introduction:** Adequate physical activity (PA) has the potential to improve quality of life (Castellano et al. 2013) and has been shown to positively influence or even reduce late effects in childhood cancer survivors (Kelly 2011). The most influencing sports structures for developing a long-term active lifestyle in childhood are physical education at school (PES), active participation in sports clubs and PA during leisure time. Especially in Germany, it has not yet been sufficiently examined whether young cancer survivors return to these sports structures following treatment. Therefore, we aimed at analyzing the survivors' status of integration into PES, club and leisure time sports activities.

**Materials and Methods:** Data was collected using the standardized PA questionnaire of the German Health Interview and Examination Survey for Children and Adolescents (KiGGS) to enable comparison with a healthy reference population. This tool was supplemented by questions regarding aspects of disease and therapy to identify barriers concerning participation. The survey was conducted in two steps: 1) with all available childhood cancer survivors over a period of three months to gain an overall impression of integration, 2) with childhood cancer survivors up to three years post-diagnosis in order to specify the decisive phase for reintegration. All participants had finished acute cancer treatment.

**Results:** In step one, 85 survivors (aged  $13.5 \pm 3.7$  years; 56% male; mixed entities)  $4.6 \pm 3.6$  years post-diagnosis were included. Overall, the status of integration into club (53%) and leisure time sports (68%) was comparable to reference data. However, 25% of the sample was exempted from PES.

In step two, 114 participants (aged  $13.5 \pm 4.0$  years; 61% male; mixed entities)  $1.7 \pm 1.0$  years post-diagnosis were questioned. Although 72% of the group desired participation in PES, 38% were not participating to full extent (17% no participation, 21% partly participation). Identified barriers included personal (physical/psychosocial), social (parents/classmates) and structural reasons (teacher/curriculum). Engagement in club (28%) and leisure time sports (37%) was considerably reduced in this group close to end of therapy compared to the reference population (58%/61%). Within the groups, most problems were revealed in brain and bone tumor patients. Both subgroups showed a status of integration below-average and the highest proportion of exemption from PES.

**Conclusion:** Generally, these results show that survivors can develop an active lifestyle and manage reintegration in sports structures. However, too many former patients do not return to previous or desirably sports activities. Especially brain and bone tumor patients seem to be disadvantaged and need individually-tailored and entity-specific support.

Furthermore, in spite of the children's motivation a high percentage is not participating in PES. Initial attempts of a reintegration program at our department showed that barriers can be successfully overcome by communication, professional advice and support provided for patients, teachers, coaches and parents. Thus, it might be possible to achieve early reintegration in sports structures and promote a long-term active lifestyle in childhood cancer survivors.

### **International project on prospective analysis of radiation toxicity in childhood and adolescence (IPPARCA): patients will profit from results**

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Late effects after radiotherapy in childhood and adolescence have mainly been characterized without detailed information regarding radiation volumes and doses at the organs at RiSK. To overcome these limitations, the Registry for the evaluation of late side effects after radiotherapy for malignant diseases in childhood and adolescence (RiSK) was established in Germany and received financial support of the Deutsche Kinderkrebsstiftung since 2004. A further cooperation between the German RiSK group and the "Swedish Paediatric Radiation Oncology Group" (SPROG) has been evolved. In May 2008, RiSK and the SPROG decided to enter into an agreement of cooperation in the "International Project on Prospective Analysis of radiation toxicity in Childhood and Adolescence (IPPARCA)". The Swedish group gained a national financial support from the "Swedish Children's Cancer Foundation" to establish structures similar to those in Germany in order to establish a prospective trial on side effects after radiotherapy in children and adults. In 2014, Norway signed the contract.

Analyses regarding the pattern of acute toxicity as well as late effects (kidney, liver, thyroid gland, head and neck) revealed relatively low incidences of high grade toxicities. Current analyses focus on acute and late toxicity after radiotherapy of the lung and high grade acute toxicity. RiSK will be able to provide more detailed data regarding dose-volume-effect relations of radiation-associated side effects on paediatric oncology patients in the future with prolongation of the follow-up time and a further patient recruitment to obtain higher patient numbers in subgroups and regarding new techniques (Intensified Modulated Radiotherapy (IMRT), protons). These data offer new concepts in optimization of therapies and of estimation of prognosis.

This month, we started to offer a counselling proposal for cancer survivors, their proxies and treating physicians in cooperation with the Network oncological advisory service (NOF), also located at the Medical School in Hannover. A planned new functionality of direct online data entry at the pediatric radiooncological trial sites will support the data entry process, accelerate further data handling and analysis and will offer the possibility to extend patient recruitment to other European countries.

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**Background:** Risk-based lifelong follow-up care for childhood cancer survivors is important to detect and treat late effects. Several guidelines and models of follow-up care have been developed. During childhood and adolescence parents have an important influence on follow-up attendance of their child. Although different models of care have been described, no studies have evaluated parents' preferences on follow-up care organization. We aimed to investigate 1) organizational aspects of follow-up care that matter to parents; and 2) parents' preferences for specialists involved in follow-up and different models of care.

**Methods:** As part of the Swiss Childhood Cancer Survivor Study (SCCSS) we sent a follow-up questionnaire to parents of childhood cancer survivors who had previously completed the baseline questionnaire and whose child survived  $\geq 5$  years after diagnosis and was aged 11–17 years at study. We assessed reasons why follow-up is important to parents; what organizational and content aspects should be included in follow-up; parents' preferences for specialists involved; preferences for models of care for long-term follow-up (all on a scale 1–4); and socio-demographic information. Proposed models of care were: telephone or questionnaire, general practitioner (GP), paediatric oncologist, medical oncologist or multidisciplinary team. Clinical data came from the Swiss Childhood Cancer Registry.

**Results:** Of 309 eligible parents 189 responded (67%). Parents valued clinical reasons (mean=3.75, SD=0.33) for follow-up care higher than supportive reasons (mean=3.11, SD=0.58;  $p<0.001$ ). They valued competent staff (mean=3.85, SD=0.37), being taken seriously (mean=3.86, SD=0.35), opportunities to check for cancer recurrence (mean=3.89, SD=0.39), late effects screening (mean=3.79, SD=0.45), and reassurance about child's health (mean=3.78, SD=0.47). The preferred specialists were paediatric oncologists (mean=3.73, SD=0.68) and GPs (mean=3.28, SD=0.89). Parents valued follow-up care by the paediatric oncologist (mean=3.49, SD=0.80) or by a multidisciplinary team (mean=3.14, SD=0.06) highest and favoured follow-up by telephone or questionnaire least (mean=1.80, SD=0.81;  $p<0.001$ ).

**Conclusion:** We showed that parents value clinic based follow-up care by paediatric oncologists or a multidisciplinary team to reassure them that their child is healthy. Parents' preferences for the organization of long-term follow-up care for young childhood cancer survivors should be taken into account when setting up new follow-up programmes.

Following a 2014 September meeting at the Romanian Black Sea coast, where 30 young people from 5 European countries met together and for the first time discussed the need of a European wide youth cancer network, in December the Romanian cancer charity, Little People, hosted the first official Youth Cancer Europe Open Space Summit in Bucharest, where 15 nations were represented by survivors and youth cancer advocates.

As one of the key outcomes of the December 2014 Open Space Summit, where common concerns were reiterated, three main topics were identified for further follow up by the group of survivors: 1. Age appropriate and equally accessible cancer treatment across Europe, 2. Long-term follow up and quality of survivorship and 3. Political and regulatory representation of young people with cancer.

As the first specific output by the newly formed European youth group, point 3: Political and regulatory representation of youth with cancer, was identified as the primary topic of further discussion. In order for young cancer survivors to deliver important outcomes and become a relevant stakeholder at political and regulatory level, young people themselves need to develop capacity in representation, networking and lobbying and learn skills in advocacy to become more capable of using their leverage with public authorities, legislators and professional organisations.

To this end, the Youth Cancer Europe network called a meeting in August 2015 in Cluj-Napoca, adjoining the Romanian survivorship community's annual general assembly and teaming up with the local Center of Resources for Public Participation, and organised an Advocacy Masterclass over two days in support of youth cancer initiatives at national or international levels, to showcase and build on existing experience and best practice, for young people to build affiliations and most importantly build skills in youth cancer survivors.

The presentation will discuss the outcomes of the above-described Advocacy Masterclass for survivors in August. (N=70 – registration is still ongoing) Since the meeting has not happened yet, we can't give a more detailed abstract, but I wanted to reserve space for it on the Vienna meeting. Thank you for the consideration!

**Background:** In Lower Saxony, a federal state in the north-west of Germany, about 180 children and adolescents develop an oncological disease every year. The significantly improved survival rates in paediatric oncology allow the majority of patients to refocus on the pursuit of their private lives after completion of therapy.

Whilst the support of the patients during therapy is well organised, the availability of long-term counselling, specifically psychosocial long-term care, shows significant deficits. To improve this situation, the project “network oncological specialist advisory service”, in short “NOF” was implemented.

**Methods:** The project focusses on the documentation of existing medical, psychosocial and other support structures for long-term after-care for paediatric cancer patients. The aim is the build-up of necessary support structures in order for the patients to cope with everyday life after therapy.

The project is accompanied by a scientific analysis of the demand for support. Therefore qualitative guided interviews were carried out with former patients. Based on the interview results, a questionnaire survey will be conducted in order to analyse comprehensively the support requirements stated by them.

**Results:** Since its start in 11/2013 the NOF is active in research and interconnection of existing local, regional and nationwide oncological long-term care projects and support facilities.

A hotline was installed in 01/2014, offering the coordination of integral support for the former patients in the areas of medical, psychosocial, legal and social issues. Since activation of this hotline, the NOF successfully dealt with 72 of 81 individual requests. The identified core problems focus on the areas of education and career choices, the application of compensatory measures in school, the communication with general practitioners, the lack of psychological support, and information deficits in the areas of gynaecology, cardiology and endocrinology.

Between 12/2014 and 03/2015, 13 guided interviews were conducted and analysed using qualitative content analysis. Identified core categories of concern include:

- psychosocial problems,
- problems with the return to everyday life after acute therapy,
- coming to terms with being a (former) cancer patient,
- coming to terms with the form of medical treatment,
- follow-up support of professionals and in health care
- information flow
- long-term consequences of cancer and treatment

**Discussion:** The results of the NOF activities show, that a considerable number of former paediatric cancer patients perceive their lives as being severely impaired after the disease/therapy.

This seems to be primarily caused by a lack of contact and information flow, but also by the general confusion of the patients social surroundings and the health care system how to respond adequately to the patients individual challenges.

The former patients shift between the desire to be cured / healthy and the tension of dealing with the often chronic consequences of the disease / the recognition of their special needs as former patients. This can lead to the development of complex, psychosocial problems and other needs for specific support.

Introduction: Transition from paediatric to adult care is an important step for many childhood cancer survivors when they reach adult age. Various barriers have been recognised on the patient/survivor, provider and health system level. Knowledge about cancer, treatment and the adult care system, or readiness to assume responsibility have been acknowledged as important aspects for successful transition. Other psychological aspects might be important barriers to successful transition. We aimed to summarise the findings on psychological obstacles preventing successful transition to adult care in childhood cancer survivors

Methods and Results: We searched PubMed without language restrictions including all years up to 5 August 2015. We used search terms addressing childhood cancer, transition, and psychosocial aspects. We found 213 articles fitting the search terms, and excluded 175 and 38 articles after screening titles/abstracts and full texts, respectively. We extracted the information on psychological obstacles for transition from 8 articles (6 qualitative and 2 mixed-methods studies; 5 from the USA and 3 from Canada). The following obstacles for successful transition to adult care were reported:

- 1) Worry about leaving paediatric oncology: Survivors reporting higher worries, were less likely to begin making changes towards transition.
- 2) Avoidance of bad memories: Traumatic experiences related to illness experience, difficulty going back to hospital, traumatic experience for the family might all decrease likelihood of successful transition.
- 3) Wanting to be normal and leaving cancer behind: Not understanding cancer as part of current life might decrease likelihood of attendance to follow-up, cancer identity or cancer as on-going part of life, feeling healthy with minimal risk for late effects.
- 4) Fear and anxiety of adverse outcomes might hinder attendance. In contrast due diligence and gratitude, and wanting to be sure to stay healthy and disease-free will improve likelihood of successful transition.
- 5) Lack of self-advocacy and communication skills. Survivors need to advocate for themselves and be able to clearly communicate with the adult health care team.

Conclusion: Psychological aspects are of great importance in addition to knowledge, availability of a competent adult care provider, insurance and other system related aspects. Transition programmes should aim to address psychological obstacles at an early stage of the transition process to guide the survivor successfully to long-term follow-up in adult care.



**Screening for cognitive deficit in 8 to 14 year old children with cerebellar tumours using self-report measures of executive and behavioural functioning and health-related quality of life****K. S. Bull<sup>1</sup>, C. Liossi<sup>1</sup>, J. L. Peacock<sup>2,3</sup>, H. Yuen<sup>1</sup>, C. R. Kennedy<sup>1,4</sup>**

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**Introduction:** We aimed to identify a brief screening measure to detect cognitive deficit in children treated for cerebellar tumours that would be useful in clinical practice.

**Materials and Methods:** 72 children aged 8–14 years and within three years post-diagnosis for standard risk medulloblastoma (n=37) or low grade cerebellar astrocytoma (n=35) and 38 in a non-tumour group were assessed using teacher-, parent-, and child-report of the Behaviour Rating Inventory of Executive Function (BRIEF), Strengths and Difficulties Questionnaire (SDQ), and Pediatric Quality of Life Inventory (PedsQL). The accuracy of these scores as a screen for a full scale Intelligence Quotient (FSIQ) <80 on the Wechsler Intelligence Scale for Children (WISC®-IV UK) was assessed using their receiver operating characteristic (ROC) curves.

**Results:** The questionnaires with the highest areas under the ROC curves were the child- and parent-report PedsQL and the teacher-report BRIEF and SDQ. At optimal cut-off scores, their sensitivities (95%CI) to cases of FSIQ<80 were 84 (60–96)%, 65 (41–84)%, 79 (54–93)%, and 84 (60–96)% and their specificities (95%CI) were 79 (68–86)%, 87 (77–93)%, 77 (66–86)%, and 71 (64–84)% respectively. All cases of FSIQ<80 screened positive on either teacher-report SDQ or self-report PedsQL.

**Conclusions:** The PedsQL child- and parent-report and the teacher-report BRIEF and SDQ have moderately good accuracy to discriminate between children with and without a FSIQ<80. The PedsQL could be used in a clinical setting, and the BRIEF and SDQ in an educational setting, to screen for cases with FSIQ<80 in children treated for brain tumours.

Within the last decades late effects after treatment for pediatric CNS-tumors have become a paramount issue. Survivors are especially at high risk for developing functional deficits. Consequently, restrictions in participation in everyday life may occur. Besides illness- and treatment-specific aspects, especially early school (re-)integration turned out to be a good predictor of a high participation level. However, only in the past few years, appropriate outcome measures were used, i. e. neurocognitive functions like attention, memory or executive functions were included in the studies. Often a discrepancy between the scores on psychological tests and observations of patients, parents and teachers in everyday life occurred, which led to misunderstandings in terms of rehabilitative interventions.

Therefore, the present study aimed at filling the information gap between neurocognitive test results and individual perceptions of the patients' possibilities to participate. In order to investigate those functional deficits that lead to participation restrictions in everyday life, the "Participation Scales 24/7" were developed. This set of questionnaires is supposed to collect data from different perspectives and, thereby, includes the views of students, parents and teachers on the individual participation possibilities of a chronically ill child or adolescent. The WHO-International Classification of Functioning, Disability and Health offered the framework for the construction of single items of the questionnaires.

After the development of the Participation Scales, altogether 1022 children were included in this study. 134 CNS-tumor patients were compared to a group of 45 non-CNS cancer patients, 258 patients with different chronic conditions as well as 585 healthy controls. For sampling, a multi-center-approach was used, including different hospitals in Austria and Germany. The test-theoretical analyses revealed that the questionnaires are a reliable and valid tool for investigating participation.

According to the results, CNS-tumor patients are at a very high risk for developing participation restrictions in the course of the illness. Illness- and treatment-related factors, e.g. the neurological or endocrinological condition of the patients, do influence functioning and participation to a large extent. These restrictions can easily be detected by the instrument developed in this study. The benefit of the Participation Scales is the possibility to directly target the subsequent interventions to these participation restrictions. Moreover, by using this instrument, not only deficits but also strengths, which may be used for compensation, can easily be identified. By including different perspectives (patients, parents and teachers), additional valuable information can be used for planning neuropsychosocial treatment. Furthermore, the Participation Scales may be used as an endpoint in CNS-tumor therapy optimizing trials, whenever survival rates of two different treatments can be regarded as equal. Then, fewer functional deficits and a higher degree of participation may be the outcome.

**Introduction:** Endocrine complications are among the most commonly diagnosed chronic conditions in childhood cancer survivors (CCS). It is established that the survivors, exposed to cranial radiotherapy are at particularly high risk of endocrine complications, but also metabolic complication and cognitive dysfunction. The challenge is to guide the survivors through the potential late complications and offer care designed to meet their specific needs to optimising their quality of life. Survivors' opinions of the quality of care they receive is an important endpoint when evaluating the healthcare. With the questionnaire "Quality from the patients' perspective" (QPP) we are able to analyse areas of satisfaction and areas in need for improvement at the clinic. Thus, the aim of the present study was to investigate CCS survivors perceptions of quality of long-term follow-up care and currently identify areas for improvement at an endocrine clinic at a Swedish University Hospital.

**Method:** The Late effect clinic of the department of oncology at the University hospital in Lund, Sweden refer childhood cancer survivors treated with cranial radiotherapy, total body irradiation and surgery to the endocrine clinic. At the endocrine clinic the survivors are tested for endocrine disorders and the most frequent conditions are pituitary insufficiency, hypogonadism and hypothyroidism. The survivors are provided with an endocrine nurse, specialized in late complications after childhood cancer, playing a key role in the care of the survivors. Quality of care at the endocrine clinic was assessed using the questionnaire: Quality from the patients perspectives (QPP). With the questionnaire we are able to analyse areas of satisfaction and areas in need for improvement at the clinic. 43 survivors treated for childhood ALL, craniopharyngeoma and CNS-tumours were included in the study. The age at diagnosis was 4.6 (1–17) and the follow-up were 32 (14–42) years after diagnosis.

**Results:** The survivors highest positive ratings were for the following items: "The nurses were honest to my questions", "My relatives were countered correct", "I received treatment without waiting", "The nurses showed engagement" and "The nurses countered me with positive mood". Inadequate quality and in need for improvement were identified for the following items: "Information about self care", "Information about the results of the examinations", "The care was customized to my needs, rather than the caregivers routines", "The physicians understand my personal situation", "I received correct diagnosis" and "Nice atmosphere at the clinic".

**Conclusion:** The survivors were satisfied with the staff-patient interaction in terms of being encountered correct, honest and with engagement. On the other side, areas in need for improvement were to offer the care with a person-centered approach, in particular regarding providing the survivors with information and to meet the specific needs of the survivors.

**Background:** Previous research has shown that adult childhood cancer survivors (CCSs) have many worries arising from their childhood cancer. We re-analyzed data from a previous employment survey(1) of CCSs in order to identify specific worries and their associated factors.

**Methods:** The participants were selected from the membership directory of Heart Link mutual-aid health insurance, and recruited by the Childhood Cancer Patients' Network. We conducted a cross-sectional survey (a self-rated questionnaire on employment) via postal-mail or an e-mail with a website link. We examined the association between characteristics of CCSs and their specific worries. The adjusted odds ratios for the factors with an interested worry were estimated using logistic regression analysis.

**Results:** A total of 239 questionnaires were included in this study. The median age of the participants was 24 years (range: 16–42 years). More than half of the CCSs had previous hematological cancers, and approximately 15% each had been treated for brain tumors and solid cancers. The most common worries were health-related problems (50%) and employment issues (40%), followed by character and life (23%) and self-appearance (20%). Fifty (21%) CCSs responded with no specific worries. CCS worries were not found to be associated with age at diagnosis or follow-up, gender, educational status, or marital status. Employment-related worries were associated with economic status, disability qualification, and employment status. Late effects was the only remaining factor significantly associated with health-related worries (OR = 2.31; 95% confidence interval [CI]: 1.21–4.39 in Table1) and with employment worries (OR = 4.03; 95% CI: 1.81–8.96 in Table 2) after adjusting other confounding factors. Late effects had the most significant association (OR = 5.93; 95% CI: 2.27–15.5), and good economic status was negatively associated with concern regarding self-appearance (OR = 0.41; 95% CI: 0.17–0.99).

**Discussion:** A previous Childhood Cancer Survivor Study reported that higher levels of anxiety among CCSs were associated with female sex, low household income, lower educational attainment, unmarried status, unemployment in the previous 12 months, and poor physical health status(2). Between 2007 and 2009, we performed another cross-sectional survey in Japan using self-rating questionnaires(3). We reported that lower scores for physical function correlated with recurrence and late effects, and poor scores for general health also correlated with late effects(3). In the present study, we confirmed that the presence of late effects was the most significant factor associated with worries of CCSs affecting quality of life.

**Conclusions:** Worries of CCSs were strongly influenced by the presence of late effects. No significant association was found between CCS worries and gender, age, or educational achievements. Economic status and disability qualification was associated with specific worries.

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350 children and teenagers are diagnosed with cancer every year in the Czech Republic. Too many of them fail to recognize the cancer symptoms early. MY NEW LIFE, the charity project under Haima CZ, association of childhood cancer survivors and parents of childhood cancer survivors, is involved in awareness activities through education sessions in elementary schools and high schools. The lessons are designed for students from 8 to 18 years of age with the main goal of raising awareness of childhood cancer in children and young people. The sessions are typically 45–60 minutes long and are tailored to the audience based on their age. The sessions are led by children oncologist and a cancer survivor. The doctor introduces basic facts and figures concerning children oncology. The cancer survivor talks about his story. The topics discussed include early warning signs of cancer, children and teenage cancer statistics, types of treatment and ideas on ways of spreading childhood cancer awareness. These sessions also encourage children and teenagers to take responsibility for their health and make positive lifestyle decisions. They remove the taboo surrounding the topic of cancer and encourage young people to talk openly about the disease. Within the last 4 years (2012 – 2015) we provided education sessions in more than 30 schools to more than 2,5 thousand students all over the Czech Republic. Our education sessions leave a great impact on our students. It was therefore no surprise, that the volunteers, who worked on the production team of the first Childhood Cancer Day in the Czech Republic in February 2015, were mostly students recruited from the education sessions.

The role of the play specialist within the Retinoblastoma team is to provide psychosocial support for children diagnosed with retinoblastoma and their families, throughout the patient journey from diagnosis to long term follow-up. The residential weekend, which is the focus of this study, is part of a wider, comprehensive, peer-support programme.

We provided a residential weekend for children aged 9–14 years who had previously received treatment for retinoblastoma. The cohort consisted of 14 young people aged 9–14, 10 of whom had one artificial eye and one having two artificial eyes. Visual acuity ranged from normal vision to blind. The choice of activities took into account the visual impairments within the group and age and abilities of the young people. The centre was well equipped for a wide range of needs. The aim of the weekend was to encourage the young people to enhance their self-confidence and self-esteem by; attempting 'adventurous' activities, taking part in specific workshops and discovering their strengths and inner qualities. The young people had opportunities to share their life experiences and ask questions about their hospital experiences should they wish to.

The weekend consisted of; centre run activities (led by qualified members of staff at the residential centre), workshop/ activities led by the hospital staff attending, and social time in which the young people were encouraged to facilitate their own activities and social interaction. The delivery style had the aim of allowing the young people to feel relaxed and comfortable in participating in general conversation. Although not a specific aim, with retinoblastoma being a common factor, discussions regarding the condition were both intentional and incidental.

The young people had the opportunity to enhance their independence by being away from their home environment in order to increase their social skills. It also allowed them to meet young people affected by the same rare condition and facing similar life challenges

The tool used to capture the young person's self-evaluation was called the "blob" tree (1), which is a qualitative assessment tool comprising 6 questions, asked at both the start and the end of the weekend, coupled with practical modelling and positioning of a plasticine person on a symbolic tree. The initial findings show increased confidence when meeting new people and in discussing retinoblastoma, and acquisition of new coping strategies through shared learning from life experiences. Parental and young person evaluation of the weekend will be presented. This residential has demonstrated the valuable role peer-support can play in the psychosocial development of young people living with the late effects of retinoblastoma. Techniques used to improve psychosocial wellbeing may have considerable value within the wider population of long term survivors of childhood cancer.

(1) The Big Book of Blobs, Pip Wilson and Ian Long, 2007

Introduction: Within the pediatric oncology, it is known that around 2/3 of former childhood cancer patients have long-term consequences and some of them have a risk for developing a second cancer. So far in Austria doesn't exist an adequate and coordinated transition from pediatrics to adult medicine for all survivors.

Materials and Methods: Two years ago, the Austrian survivors group started a working group together with the parent organisation "Österreichische Kinder-Krebs-Hilfe" and some paediatric oncologists, who are specialists in late-effects and long-term follow-up. Because of the dissatisfaction and the fact that many survivors are lost in follow-up this working group started a brainstorming process, where survivors could mention their wishes towards the long-term follow-up.

Results: After half a year exchange within the working group members, the need of a special transition model was obvious. The outcome was the piloting of a special transition model called "ZONE", which is the abbreviation for Centre for Oncological follow-up of adult (who has as a child or teenager an oncological disease). The idea of this transition model ZONE is to have an adequate and individual transfer from the pediatric oncology/hematology to disciplines in adult medicine. It should be a competence center for oncology (medical and psycho-social) long-term follow-up and a contact point for former patients and for their family doctors. One important aim is to raise awareness about the importance of adequate follow-up in Austria. Furthermore this transition model should support and promote the research of long-term consequences for pediatric oncology diseases.

Conclusion: The establishment of this special transition model will close the gap within the health care system in Austria and will offer a coordinated, high quality follow-up for Survivors. Making this come true within the Austrian health care system is only possible with the collaboration of survivors, parents and medical professionals.

**Background:** Survival of children treated for cancer is constantly improving, late sequelae of childhood cancer treatment being of major concern. Slovenia is small country with 2 million inhabitants. Fifty children under 16 years are diagnosed with cancer every year. All of them are treated at Hemato-oncological department of University Children's hospital in Ljubljana and get radiotherapy treatment at the Institute of Oncology Ljubljana. The Slovenian Cancer Registry was established in the early 1950 and is based on the obligatory registration of all cancer patients in Slovenia.

**Methods:** All childhood cancer survivors are included in a program in which pediatricians, "adult" radiation oncologists, involved in radiotherapy treatment of paediatric patients, and other specialists join in the follow-up of late sequelae. This program is financed by the Ministry of science since 1986. When the childhood cancer survivor reaches the age of 18 years and is at least 3 years post treatment, he or she is eligible for follow-up at the Institute of Oncology in Ljubljana. There, a team consisted of adult radiation oncologists and nurse-coordinator, in cooperation with medical doctors of different specialties, continues follow-up for life. More than 900 patients are being regularly followed at present. We are looking for late sequelae of treatment, with specific research projects concerning organ deficits such as cardiac, pulmonary, renal, endocrine, hepatic etc., second primary neoplasms (screening diagnostic tests for early detection), intellectual abilities and psychosocial status. We offer psychological support to every survivor in the form of monthly meetings in groups of 10–12 young adults, guided by psychologist.

**Results:** From 1957 to 2004 1975 children less than 16 years of age were treated for cancer in Slovenia, 885 of them are alive, 799 older than 18 years, at least 3 years post treatment. 771 patients were evaluated. We found mild somatic sequelae in 31% of patients, moderate in 22%, serious in 13%, 4% died of second primary neoplasms and late somatic sequelae. Incidence of second primary neoplasms was 13% 25 years after diagnosis of primary neoplasm. Emotional disorders were found in 83% of patients.

**Conclusions:** incidence of late sequelae after childhood cancer treatment in our country is high and comparable with those reported in the literature. Regular follow-up of long-term survivors of childhood cancer is mandatory. An integrated program, such as ours in Slovenia, can be used as a model for organizing the long-term follow-up of late sequelae after childhood cancer treatment.



At The Royal Marsden Hospital NHS Foundation Trust all children and young people (CYP) 2 to 5 years following the completion of their cancer treatment are referred to the long term follow up (LTFU) service. Between the ages of 16 and 25 they are seen in the teenage and young adult (TYA) clinic. When ready they are transferred to the Pan London adult LTFU service based at University College London or discharged to supported self management.

#### Reviewing the Service

Theoretically we had all the components for a model transition clinic. A clinic specifically for TYA's well staffed with a clinical nurse specialist, a nurse consultant in teenage cancer, a paediatric oncologist and an adult endocrinologist. All patients provided on entry with an individualised treatment summary and long term follow up care plan. The clinic experience however could be stressful for patients and staff. Large volumes of information needed to be discussed. Some patients became upset on receiving their written summary and care plan, seemingly hearing the information contained within them for the first time. TYAs were encouraged to come in for some of the appointment alone but many parents were anxious about this and reluctant for it to happen.

#### Identifying the changes required

To address the challenges we used 'Benchmarks for transition' (Gibson et al 2014) to identify areas for improvement. Recognising transition should not be a one-off event but a process we started transition in the children's clinic. A nurse clinic was established enabling more time for communication directly with the young person because simply being present in the clinic room does not always mean the child is engaged. We began working earlier with the whole family to help them understand the value of the young person actively managing their own health needs.

We decided to implement Ready, Steady, Go, a framework for transition involving a series of questionnaires that are completed by the child and parent/career (Nagra et al 2014). It is quick and easy to use, offers a structured approach to transition, includes parents, and is patient led. It is designed to be generic so is suitable for our patients with their wide range of cancers and treatments. It provides an assessment of 'readiness' enabling concerns to be addressed early, thus helping reduce the anxiety felt by some families moving on from children's services.

This presentation aims to demonstrate that generic transition tools can be successfully applied to children's cancer follow up. Young people and families at the Royal Marsden Hospital have responded well to the introduction of Ready, Steady Go. We hope we will see this reflected when we repeat our patient experience audit next year. The changes have generated a lot of interest amongst our acute colleagues. We see a time when Ready, Steady Go will be implemented across the whole of CYP cancer services.

## Long term follow up consultation after childhood cancer in France (Rhône-Alpes region): the voices of adults and their general practitioners

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In France, about 50 000 adults are potentially concern by late effects after childhood cancer, but not many know the types of anti-cancer treatments that they received (Berger C et al. Bull Cancer. 2015). Models of care for this population are various (Heirs M et al. Pediatr Blood Cancer. 2013, Singer S et al Pediatr Blood Cancer. 2013) : patient driven, general practitioner (GP)-led, nurse-led, medical specialist-led, in a long term follow-up clinic or in the community.

**Objectives:** to evaluate the level of satisfaction with their care of 150 adults cured of childhood cancer and their general practitioners .

**Aims and methods:** the long-term follow-up study in oncology (SALTO) is an ongoing prospective cohort study of childhood cancer survivors (except leukemia) diagnosed between 1987 and 1992 in the Rhône-Alpes and Auvergne regions of France (n= 508). After agreement, 150 respondents consulted with a pediatric oncologist and an internist. Additional tests scheduled according to recommendations after chemotherapy and and/or radiotherapy were proposed (and specific organ cards founded in <http://sfce.sfpediatric.com/page/le-suivi-à-long-terme> were given), along with advice for prevention and follow-up. A summary of the oncologist consultation was sent to the survivor and GP Two years after the initial consultation, a satisfaction questionnaire was sent to the adults and there GP.

**Results:** The vast majority (89%) of survivors participating (response rate 80%) were satisfied with the consultation. The majority of the participants (51 %) would again like to benefit from a SALTO consultation, 28 % would not wish it. Concerning the recommendations cards given at the end of the consultation, 96% of the the adults who received at least one card read them, most ((86%) found them useful and 71 % followed the recommandations. After the SALTO consultation, 32% said that they had changed their medical care with regular follow-ups with a GP, or specialist, or internist. .

Of 137 surveys sent to the GPs 77% returned to us. Most GPs were poorly informed about the long-term complications after chemotherapy (77%). More than half of them (59 %) did not know treatments received during childhood cancer and 70 % made no follow-up before the LTFU consultation.

Report of the SALTO consultation was found useful by 61 % of these doctors, 82 % found the mixed consultation oncologist pediatrician / internist useful and 88 % found helping to have a hospital contact for this specific population.

**Conclusion:** LTFU specialist consultations with summaries to survivors and GPs are acceptable to both. Majority of internists are willing to follow these adults in collaboration with pediatric oncologist. We propose in the future at least one common consultation and a regular follow-up by internist for high risk survivors.

**Background:** Transition from child to adult health care for young people with long-term conditions is currently a 'hot topic' within UK health services as well as elsewhere in Europe. In spite of the growing need, there continues to be a lack of empirical evidence and solid evidence for what works in practice is lacking (Crowley et al, 2011). The implementation of transitional care remains a challenge. Lack of 'being prepared' was a main finding from young people and parents reported by the UK Care Quality Commission (CQC) in 2014. Here only 54% of young people described preparation for transition that had enabled them to be involved in the process and 80% of pre-transition case notes reviewed had no transition plans for health. The CQC recommend that existing good practice guides are followed to ensure young people are properly supported through transition. Benchmarks offer a guide/standards that services can measure themselves against to see how they are doing, where they could improve and can facilitate the sharing of best practice. Establishing a good partnership between paediatric and adult providers, implicit throughout the benchmarks, has since been confirmed in an International Delphi study (Suris et al 2015).

**Aims:** To describe the development of a clinical practice-benchmark tool for transition.

**Method:** This qualitative study involved focus groups, workshops and interviews. Data were collected with young people with long-term health conditions, their parents, professionals and experts leading on transition within the UK. Transcripts were analysed using qualitative content analysis. The focus was to develop an increased understanding of transition, from multiple perspectives, and to describe what strategies and resources might be required to facilitate transition with the aim of developing a benchmark tool.

**Results:** For young people and their parents/carers to experience timely and effective transition, eight factors and their associated indicators of best practice statements have been developed. In order to ensure the benchmarks reflected their needs and preferences, young people and parents led on the selection of factors, practice statements and indicators of best practice. Communication, co-ordination, gradual transition and support to manage their health condition as an adult were paramount for them. The tool was distributed to a range of professionals across the UK for comment and subsequently refined to produce the current benchmarks. The benchmarks have subsequently been piloted with five sites in the UK.

**Conclusion:** The need for change in order to best meet the needs of young people, and parents, during transition is evident. The benchmarks indicate young peoples' and parents' needs and preferences regarding transition to adult care. Working with sites to pilot the benchmarks and gain feedback on their use has demonstrated their usefulness in facilitating dialogue within teams about improving transition and in the sharing of good practice. This paper will present the benchmarks for transition and describe the process of their development, and refinement.

## **No evidence for longterm chemotherapy-induced neurotoxicity for pediatric ALL patients: A prospective study**

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**Introduction:** Survival rates of pediatric Acute Lymphoblastic Leukemia (ALL) patients have consistently been increasing during the last decades. Given that, since the end of the 80ths, we did not use any radiotherapy for central nervous prophylaxis in our frontline therapy anymore, and our treatment became only chemotherapy based, chemotherapy-induced side effects such as cognitive decline stay important to assess. Although chemotherapy-induced neurotoxicity was demonstrated in several cross-sectional studies so far, the number of large follow-up studies remains limited. Still, to investigate neurotoxic effects, a longitudinal design is most suitable.

**Materials & methods:** In this study we evaluated intellectual development of 94 Flemish pediatric ALL patients, standard risk, diagnosed between 1990 and 1997, with a prospective design. Median age was 4.4 years (2.0–11.9 yrs). All patients were treated according to the EORTC 58881 protocol without cranial irradiation. Intellectual functioning was assessed with the WISC-R at three time points since diagnosis with an interval of three years; but age at baseline assessment was minimal 6 years. Hence, patients who were diagnosed before 6 years old, had already received chemotherapy by the time the first assessment took place. In a fixed effects repeated measures regression analysis the effect of gender, disease risk, education of the parents, age at diagnosis, IQ subscale (verbal (VIQ) vs performal (PIQ) intelligence), and test moment were included as predictors for IQ, to address potential delay in neurocognitive development.

**Results:** IQ scores did not decline over time. VIQ scores were lower at baseline but hardly changed over time (i.e. test moment), whereas PIQ scores increased over time such that the difference with VIQ scores disappeared at the third assessment. IQ scores were not predicted by gender ( $p=.78$ ) nor by disease risk ( $p=.49$ ), but were significantly higher when at least one parent had followed higher education ( $p<.0001$ ). What is more, a lower age at diagnosis ( $p=.0009$ ) predicted lower IQ scores at baseline. More specifically, there was a significant interaction effect of IQ subscale and test moment ( $p=.0079$ ).

**Conclusions:** Given that IQ scores did not decline, nor did disease risk relate to cognition, our findings suggest the absence of longterm neurotoxicity due to the specific chemotherapeutics that were administered. Lower IQ scores for patients who were diagnosed at younger ages however, highlight the stronger impact of the disease and/or treatment at younger age. Given that PIQ was most explicitly lowered at baseline, specifically for patients diagnosed at younger age, our results could suggest that performal functioning is more vulnerable to acute neurotoxicity.

**Background:** Since 1980 patients with Ewing sarcoma have been treated according to consecutive protocols of the German Society of Pediatric Oncology and Hematology (GPOH). Rising survival rates have raised the question of the quality of long-term survivorship. Objective and subjective measurement tools are used to evaluate the actual health status and daily-life activity level as an indicator for restitution of function.

**Methods:** Long-term outcome of 603 survivors of the CESS81, CESS86, EICESS92, and EURO-E.W.I.N.G.99 trials, diagnosed between 1980 and 2009, was assessed by the Toronto Extremity Salvage Score (TESS), Short-Form Health Survey (SF-36), and Brief Symptom Inventory (BSI) questionnaire scales, and by the StepWatch Activity Monitor (SAM) accelerometer device. A 1:2 non-random peer control group was selected to compare results with healthy individuals. Median age of former patients was 28.7 years, 56% were males. Median observation time was 12.9 years (range 3.7–31.2).

**Results:** Former patients were less active than the control group, contributing to a mean step count difference of 1758 steps per day (10394 vs. 12152;  $p < 0.01$ ). Negative prognostic factors were pelvic tumors (9265;  $p < 0.01$ ) and primary metastatic disease (9322;  $p < 0.05$ ). Correlations between self-reported scales and the step measurement were rather low ( $r < .30$ ), BSI somatization, anxiety and depression scales (raw values  $< 0.50$ ), and the SF-36 (Physical/Mental Component Summary scores=47.9/49.7) showed no major clinical or functional limitations. Around 15% of former patients rated their health status as less good or poor compared to 2% of the controls.

**Conclusion:** The present study comprised a follow-up period of up to 30 years after the treatment of Ewing sarcoma. Former patients seemed to return to a normal lifestyle with minor limitations. The generally positive outcome is an encouraging finding for patients with this severe disease.

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During the last years several studies were published about the health status of children born to survivors of childhood cancer from different countries, for instance Northern European Countries, United States, The Netherlands (Winther JF et al 2010, Green DM et al., 2010, Lie Fong S et al. 2010). These results are very encouraging for counselling young adults after childhood or adolescent cancer if they wish to conceive after treatment. Pelvic irradiation was found as a risk factor, there is an increased risk for low birth weight and premature birth. But no additional risk for malformations or other birth problems were found. In Germany, Austria and Switzerland most of the children are treated according to the same or very similar protocols. We got the possibility to take part in the FeCt– Multicentre Offspring Study (lead by A. Borgmann–Staudt, Berlin, Germany). In this study offsprings of former childhood cancer patients of these three countries are included. We evaluated the questionnaires sent to former patients of the St. Anna Children's Hospital, Vienna, Austria, to compare our institutional results with the results of the larger group.

Questionnaire			
<b>Questionnaires sent out</b>	471	<b>Children</b>	<b>39</b>
undeliverable	41	1 child	20
<b>Questionnaires answered</b>	<b>153 / 36%</b>	2 children	16
<b>Sex ratio</b>		3 children	2
male	83	4 children	1
female	70		

Characteristics of the offsprings			
<b>Sex ratio</b>		<b>Malformations</b>	
male	21	Frenulum of tongue	2
female	18	Umbilical hernia	1
<b>Birth weight</b>		<b>Oncological diseases</b>	
under 2500 g	0	Leukaemia	1
2500 – 3500 g	30		
over 3500 g	6	<b>Allergies</b>	
over 4000 g	3	hey fever	2
<b>Lengh at birth</b>		neurodermitis	2
between 40 and 50	19	obstructive bronchitis	4
over 50	20		
		<b>Other problems</b>	
<b>Overall health status</b>		Ametropia	8
excellent	30	Seizures under the age of 2	1
good	7	Migraine	1
moderate	2	Hyperactivity	1
poor health	0	Thyroid disease	1