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Original Article

Changes in the Patterns of Care of Central Nervous System Tumours Among 16–24 Year Olds and the Effect on Survival in Yorkshire Between 1990 and 2009

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Abstract

Aim: There is a paucity of work documenting the influence of patterns of care on survival for teenagers and young adults with primary central nervous system tumours. Therefore, the aim of this study was to undertake a detailed assessment examining any changes in the patterns of care over time and how these related to survival outcomes for 16–24 year olds diagnosed with a primary central nervous system tumour between 1990 and 2009.

Materials and methods: We used high-quality data from one population-based cancer registry in Yorkshire, UK to describe primary central nervous system tumours in teenagers and young adults (16–24 years) diagnosed between 1990 and 2009. The Birch classification scheme was used to identify differences by tumour subgroup. Incidence, patterns of care and survival trends were described using Poisson and Cox regression.

Results: There were 163 cases comprising 98 astrocytomas, 17 'other gliomas', 14 ependymomas, 11 medulloblastomas and 23 'other intracranial and intraspinal neoplasms' yielding an overall incidence of 18.1 million person-years. Care varied significantly over time and by principal treatment centre (Leeds 77%, Hull 23%), co-ordinating specialty (neurosurgery 53%, clinical oncology 22%, paediatrics 17%, other adult services 8%) and treatment received. Cox regression showed no significant difference in survival by age, gender, treatment centre, level of deprivation, year of diagnosis or co-ordinating specialty, but a significant difference by tumour grade and diagnostic group. Survival improved for all diagnostic groups except astrocytoma, although only the medulloblastoma group showed a significant change over time.

Conclusion: The lack of any significant improvement in survival over time in most diagnostic groups warrants further investigation and provides justification for a more collaborative regional approach to the care of central nervous system tumours, perhaps through the development of regional guidelines for this unique population. More detailed analysis of relapse patterns and prediagnostic symptoms would also be informative for this cohort.

Key words: Central nervous system neoplasia; patterns of care; survival; teenagers; Yorkshire; young adults

Introduction

The teenage and young adult (TYA) age group is well established as a transition group falling between the remits of traditional paediatric and adult oncology, but with unique needs [1]. Brain tumours are also unique in the extent and variety of their effect on the patient's capacity to function. Intensive support with specialist nurses, physiotherapists, speech therapists and psychologists is required to improve their quality of life and to reduce significant morbidity in survivors [2].

The UK Improving Outcomes Guidance (IOG) [3] for cancer in children and young people published in 2005 aimed to improve cancer outcomes for this population adhering closely to two key principles: to (i) treat cancers in young people as rare cancers, and (ii) use age-appropriate centrally co-ordinated models of care, including that 16–18 year olds be offered treatment at a specialist centre. Following the establishment of the adolescent (15–19 years) and young adult (20–24 years) age ranges, the IOG agreed upon the definition of TYA (or young people) as those aged 16–24 years [3,4].

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The National Health Service Specialised Commissioning Groups Children's Neurosurgical Services Review accepts that children managed through high-volume age-appropriate neurosurgical services fair better [5]. However, less than 500 paediatric central nervous system (CNS) tumours are operated on in the UK and Eire each year across 14 centres, with some older children continuing to be operated upon by adult neurosurgeons. The ongoing review will lead to a formal designation process to ensure that sustainable high-quality multidisciplinary care is delivered [6].

The distribution of tumours in this age group is unique, comprising a mix of paediatric and older adult cancers, as described by the Birch classification system for TYA CNS tumours [7]. CNS tumours account for 9% of tumours registered in the 15–24 year old age group in England [7,8]. The average annual number of newly diagnosed CNS tumours for 15–24 year olds in England is about 200, equating to an overall incidence rate of 33 per 1 000 000 person-years [9]. There is a uniform distribution for CNS tumours despite a small increase in incidence due to the 'other gliomas' group, which includes oligoastrocytoma and oligodendroglioma [7,8,10]. Improvements in survival for CNS tumours have been cited to lag behind cancers of every other primary site [11]. Clinical trial accrual among CNS tumours has been notably poor for many years, especially among 15–24 year olds [12], which may be contributing to this lack of sustained improvement in survival.

CNS tumours account for the highest rates of cancer-related mortality in this age group, in part due to the higher incidence of aggressive high-grade glioblastoma and poor outcome medulloblastoma [9,13]. International evidence shows poorer cancer survival outcomes for TYA compared with younger children (ages 0–14 years), and a notable lack of improvement in survival over the last 25 years [10] compared with older age groups (aged over 25 years) despite gradual increases in 5 year survival rates among childhood cancers.

Age and tumour characteristics (morphology, World Health Organization grade, tumour site) are well-known prognostic factors, whereas socioeconomic and geographic factors have also been shown to exhibit a small but significant role in survival [14–16].

We have published findings describing overall incidence and survival trends for CNS tumours diagnosed in young people in Yorkshire between 1990 and 2002 [16], but there is a paucity of work describing the epidemiology of cancer in relation to patterns of care for CNS tumours in TYA. For example, care can be instigated by paediatric services, with some paediatricians holding onto patients into adulthood, and others transferring to adult services between 16 and 18 years of age. The destination of referral into adult service varies, although it is usually either to adult oncology or neurology departments. Where TYA patients have been under the care of neurosurgeons or clinical oncologists, care may continue into adulthood. There are many possible permutations, and departments have long-standing arrangements in shared care through informal mechanisms.

The advent of the IOG, the formal multidisciplinary team and specialist TYA services have all strived to simplify this variation in care, as such heterogeneity has been cited as a contributing factor to the poor improvements seen in overall TYA survival. In response to the IOG and the Children's Neurosurgical Services Review, we therefore aimed to undertake a detailed assessment examining any changes in the patterns of care over time and how these related to survival outcomes for 16–24 year olds diagnosed with a primary CNS tumour between 1990 and 2009 in Yorkshire, UK. We aimed to examine a priori the clinical features of survival in relation to the principal treatment centre (PTC), the co-ordinating speciality, treatment received and deprivation.

Materials and Methods

Data were analysed for all patients diagnosed with a primary CNS tumour, aged 16–24 years old, between 1 January 1990 and 31 December 2009.

The data were extracted from the population-based Yorkshire Specialist Register of Cancer in Children and Young People (YSRCCYP), which collects information pertaining to diagnoses occurring within the current Yorkshire and Humber Strategic Health Authority on all newly diagnosed cancers in children and young adults under the age of 30 years since the mid-1970s and has full ethical approval. It contains a wide range of demographic data and detailed clinical and therapeutic information that is updated regularly through active follow-up procedures [17].

A high level of case ascertainment is ensured through abstractions of medical records throughout the Yorkshire region and regular cross-checks with the National Registry of Childhood Tumours (www.ccrg.ox.ac.uk), the Northern and Yorkshire Cancer Registry and Information Service (www.nycris.org.uk) and local neuropathology departments in Leeds and Hull. Individuals are actively followed-up every 2 years through contact with general practitioners and with hospital consultants. Pathology records are retained to ensure accurate classification of tumours. Secondary malignancies and relapses are also recorded to ensure that primary neoplasms can be identified.

Data on the YSRCCYP were further validated and augmented manually through interrogation of microfilm, paper and electronic notes, supplemented by specialist oncology database software from the PTCs in Leeds and Hull. The PTC was defined as the main centre at which each patient's overall treatment was co-ordinated after diagnosis. Data were validated for diagnostic and treatment information, and additional information was collected on the grade at diagnosis, treatment received, and co-ordinating specialty. The co-ordinating specialty indicated the speciality of the consultant co-ordinating the patient's care at the PTC. The co-ordinating specialty was not necessarily identical to treatment received; for example, a patient whose care is co-ordinated by the neurosurgeons but receives adjuvant radiotherapy.

Data were excluded if the diagnosis occurred in South Yorkshire as registrations for this part of the region only extended back to 1998 (Figure 1). Registrations were also