Subspecialisation in neurosurgery—does size matter?

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In this issue Solheim et al. [15] present data from the Norwegian Cancer Registry looking at the 5-year survival for children with brain tumours in relation to centre volume and outcome. As these authors indicate, centralisation of services is very topical at the present time. Certainly in England paediatric craniofacial services have been largely centralised for many years and there is presently a review of how best to deliver the rest of paediatric neurosurgery [14]. Likewise, in Holland there are discussions over centralisation of paediatric brain tumours (and there has already been centralisation of craniofacial and epilepsy surgery), and centralisation of paediatric neurosurgery has already begun in France [9]. In countries without socialised medicine, there is a move by insurance companies to seek treatment for their patients in accredited centres. The driver for these changes is a desire to improve the quality of care.

In their study, Solheim et al. [15] did not observe a relationship between centre volume and outcome in Norway and they raised concerns over the use of centre volume as a proxy for quality. Comparing the results from one “large” unit with those from three “small” centres, the authors found no overall difference in survival between the centres and children with primitive neuroectodermal tumour (PNET)/

medulloblastomas treated in the larger centre fared worse. The authors draw two main conclusions:

1. Although case volume is often put forward in debates on centralisation for cancer surgery, “less quantifiable factors” may be more important for the outcomes; i.e. centre volume is not a good proxy for quality
2. Due to the rarity of paediatric brain tumours (and the fact that paediatric neurosurgeons spend most of their time doing hydrocephalus work), “adult” neuro-oncology surgeons might be better equipped to undertake such surgery; i.e. the “transferable skills” component of neurosurgery is underscored

Prior to reviewing the paper by Solheim et al. [15] in more detail, let us review what is currently available in the literature, both in general terms and more specifically for paediatric brain tumours.

Volume-outcome relationships in tumour surgery

Many studies have been undertaken on this topic since the landmark paper by Luft et al. [13] in 1979, which showed an inverse relationship between surgical volume and mortality. These studies have shown that this relationship varies from one type of surgery to another. In general, the more complex the surgery, the greater the improvement in outcome with increasing volume, and the point of “plateauing” for both individuals and for institutions varies markedly depending on the condition being treated. To the proponents of centralisation, these studies confirm the intuitive belief that in surgery (as in all other walks of life) “practice makes perfect”.

Those critical of this approach argue that case volume, as a proxy for quality, is too imprecise and often concentrates

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on institutional volume, when it should focus on individual surgeon volume. This is understandable as institutional volume is often a proxy for resources and ability to set up an environment that allows the development of individual expertise. Both individual surgeon and institutional “experience” may independently influence outcome. Critics also argue that the disruption and inconvenience to the patient by centralisation is out of proportion to any potential benefits gained [10, 15]

Gruen et al. [6, 10] undertook a systematic review and meta analysis of case volume on cancer mortality and tried to answer three “policy orientated questions”:

1. What is the strength and robustness of the association, if any, between hospital or clinician case volume and patient outcomes?
2. Is the association clinically important?
3. Is there consistent evidence of a volume threshold, above which better outcomes are observed?”

The authors conclude from their analysis that studies on this topic are difficult to compare, due to considerable heterogeneity of the quality of data (particularly administrative databases) and due to a failure to account for potential confounding factors. Despite this, the authors found for many tumour types a statistically significant inverse relationship between hospital volume and peri-operative mortality”. From the studies available, the authors were unable to answer the question of whether individual surgeon volume was more or less important than institutional volume and the authors were unable to give any guidance on the “potential paradox of a low volume surgeon operating in a high volume hospital or a high volume surgeon operating in several low volume centres”.

For all tumours, the authors concluded that there is “a reduction of somewhere between 10% and 40% in mortality risk” and this “when considered on a population basis, the difference between all patients being treated in high volume centres to all patients being treated in a low volume, make up for an absolute improvement in mortality of 1–2% for most procedures”.

The third conclusion from this review was that hospital volume appeared to be associated inversely with mortality across the volume spectrum, i.e. there was no “plateau”.

The authors state: “thus, volume is only ever at best an imperfect proxy to healthcare policy. Well chosen process measures and individual risk-adjusted outcome measures may also play a role in reflecting quality of care and predicting patient outcomes”. Other outcomes, such as quality of life, may well be more important in this debate than mortality—especially from the patient and family perspective. This is particularly pertinent to paediatric central nervous system (CNS) tumours, where overall more than 75% of children will become long term survivors and

studies have shown that over half of such survivors [4, 11, 12] will be left with moderate or severe disabilities. Furthermore, unlike upper gastrointestinal (GI) surgery, CNS tumour surgery probably only carries a peri-operative mortality rate of approximately 1%—making early mortality a less useful endpoint.

Bilimoria et al. [3] took a different tack in their systematic review by looking at surgeon training, specialisation, experience and the relationship of these to the outcome from cancer surgery. Having accepted that greater volume and therefore greater experience improves outcomes, these authors were looking to see if it was possible to understand *why* patients who were treated by higher volume surgeons fared better. These authors argue that surgical specialisation (including additional fellowship training) improved clinical judgement and surgical skills in specific areas and so those undergoing it will naturally be more likely to take on a high volume practice in their chosen discipline. The authors hoped to disentangle this aspect of training versus general experience gained over time—while noting the important role that the “systems of care” (institutional “experience”) play on patient outcome. These authors found 29 published studies that met their inclusion criteria, of which 27 demonstrated that surgeons’ specialty training and increased experience resulted in better outcomes. They noted that the definition of “specialisation” ranged considerably in the studies and conclude that “it remains unclear whether additional training or a specialised practice is really what affects outcomes”. The authors remind us of the comment by Dr. Blake Cady that in the field of surgical oncology, “Biology is King; selection of cases is Queen, and the technical details of surgical procedures are the Princes and Princesses of the realm who frequently try ... to usurp the throne [but] almost always fail to overcome the powerful forces of the King and Queen” [5]. It is clear from this review that gaining surgical experience plays an important part in preparing the “queen, prince and princess”. Understanding the king (tumour biology) is part of the role of research, and again it is generally the case that the larger the centre the more likely there is to be ongoing research which may change the future direction of treatment for that tumour type.

What none of these studies can take into account is the inherent variation in surgical skills that occurs between surgeons. It will always be the case that a small volume “gifted surgeon” may well have similar (or even better) surgical results than a high volume “average surgeon”. However, what these studies show is that both surgeons will improve their outcomes with appropriate training and a job which concentrates and increases their experience.

Volume outcome studies specific to CNS tumours are limited and virtually all are published on North American

data; direct extrapolation of results to other healthcare systems is difficult but still relevant. Barker et al. [2] studied 38,000 adults undergoing craniotomy in North America and found a trend towards better outcomes, (lower mortality and lower unfavourable discharge) in larger volume centres than in smaller volume centres. These authors were unable to establish a case-load threshold. Curry et al. [7] likewise found lower mortality and adverse discharge in adults operated upon in America in high-volume providers. Albright [1] reviewed 752 children with medulloblastoma or high-grade glioma operated on by 269 neurosurgeons and 56 paediatric specialists and found the degree of resection to be higher for the paediatric subspecialists. Interestingly, the neurological morbidity after surgery was significantly lower in patients operated on by surgeons who were members of the American Society of Pediatric Neurosurgeons (ASPN) than those operated on by general neurosurgeons or neurosurgeons who were “designated” to operate on children in their hospital. Membership of the ASPN requires at least 5 years of experience in pediatric neurosurgery after the completion of residency or fellowship training and the demonstration, by submission of surgical logbooks in the 2 years preceding membership, of either (1) performance of 75% of all operations on patients younger than 18 years of age, or (2) performance of at least 125 operations per year on children younger than 12 years of age.

Smith et al. [16] looking at children with brain tumours in the United States, found a higher mortality and a greater likelihood of discharge destination other than home for the lowest volume centres and surgeons. These very low volume centres were defined as those operating on less than five paediatric brain tumours per year.

The only study we can find that compared longer-term outcomes in neuro-oncology is a study from Ontario [8], which showed that children treated for medulloblastoma at the Hospital for Sick Children in Toronto, had a 5-year relapse-free survival rate of 65% compared with 44% for those treated at other smaller centres (range 25–60%). Cases treated outside Toronto had a nearly twofold increased relative risk of death or recurrence.

Transferrable skills

Solheim et al. [15] allude to the fact that there may be transferrable skills that give adult neurosurgeons the experience and knowledge to safely treat paediatric brain tumours. As can be seen in Fig. 1, the tumour types have very little in the way of overlap; tumour location is therefore perhaps more important than tumour type. Paediatric tumours are much more likely to be found in the mid line (especially in relation to the third and fourth

ventricle) and these require specific approaches to reach them. Colloid cysts of the third ventricle make up less than 1% of adult intracranial “tumours” and are the only relatively “common” lesion at this site in adults. True fourth ventricular lesions are rarer still in the adult setting. Other tumours (e.g. pineal region tumours and intrinsic spinal cord tumours) are rare in both adults and children. The only way to increase experience in the management of any of these tumours is by centralisation. While no one would argue that certain tumours which rarely occur in children (e.g. pituitary tumours and acoustic neuromas), are probably best undertaken by “adult” neurosurgeons, the converse may also be true for “paediatric-like” tumours in adults.

The surgical experience required to undertake certain approaches is only one part of the management of children with CNS tumours. Other aspects of management—e.g. the treatment of associated hydrocephalus and the appropriate pre-operative investigations—need to be undertaken in a multidisciplinary fashion. Furthermore, the “goal of surgery” is often different for adult and paediatric patients; e.g. the adult literature appears to remain focused on obtaining a gross total resection for craniopharyngioma, while most paediatric neurosurgeons have moved away from this due to the (hypothalamic) morbidity caused by such surgery.

Lastly, we would not argue for the centralisation of paediatric tumours alone, but rather the management of all paediatric neurosurgical cases in a limited number of centres—with only true paediatric emergencies being dealt with by “adult” neurosurgeons. Apart from each of the subspeciality areas within paediatric neurosurgery benefiting from the increased experience so gained, this concentration of paediatric practice is more likely to allow the formation of a separate 24/7 on-call system for paediatric neurosurgery. The knock-on effect of increased “volume” should be improved institutional experience and memory—particularly in neuro high dependency and paediatric intensive care, neuroradiology neuropathology and neurophysiology—as well as more dedicated training in paediatric neurosurgery, research opportunities and access to non-medical specialists (paediatric neuropsychologists, nurse specialists, physiotherapist, speech and language, occupational therapists, rehabilitation, etc.). This is not to suggest that paediatric neurosurgery should break away from general neurosurgery. Far from it, it is our view that paediatric neurosurgery is and should stay a subspeciality within general neurosurgery. The question is how is the care of children with neurosurgical problems best delivered?

Analysis of the Norwegian study

An important strength of the present study is that it was carried out in the setting of the Norwegian Cancer Registry. This is a population-based national registry with verified

Fig. 1 Comparison of CNS tumour pathology between children and adults (courtesy of J. Goodden). Adult data adapted from CBTRUS (2010) Statistical report: primary brain tumors in the United States, 2004–2006: <http://www.CBTRUS-WEBREPORT-Final-3-2-10.pdf>.

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high levels of data quality and completeness, and linkage to national record systems, combined with a low level of international migration where patients would be lost to follow-up. Nevertheless, the study does suffer from several weaknesses, some of which are unavoidable:

1. The total number of patients studied, 816, may appear quite large. Spread over the 20-year study period, however, this amounts to only about 40 patients operated on per year. Of these, about half had surgery in the higher-volume centre (i.e. 20 cases per year) and the remaining half had surgery in the other three, lower-volume centres (i.e. approximately six to seven cases per year in each centre, not far above the annual number at very-low-volume centres in the study by Smith et al. [16]). Thus, in effect, the comparison is between three very-low-volume units and a single low-volume unit, rather than a high-volume centre. Moreover, the series encompassed more than 20 different tumour

types, among which even the most frequent one, pilocytic astrocytoma, accounted for a mere seven to eight cases per year nationally. It could be argued that none of the units in Norway is doing a sufficient number of operations on this disparate group of tumour types to have gained the “experience” that a high volume centre might develop. With such small numbers and no detail on the number of individual surgeons in each centre, it is quite possible that the individual surgeon volumes were similar. If surgeon volume rather than centre volume per se were a stronger determinant of outcome, it would not be surprising that no overall difference in survival was detected in this study

2. The authors acknowledge the limitations of a registry-based analysis (in particular, the lack of ability to stratify by risk factor). However, they argue that this is unlikely to have had a large effect as they have analysed the data both by centre and by patient address (equivalent to an intention-to-treat analysis in a

randomised trial). This may have held true if the numbers for each subset of tumours were sufficiently large. However there were statistically obvious imbalances, in the choroid plexus tumours group 19 patients were residents of the high volume region and only three were from the low volume regions ($p=0.004$), and there was a similar imbalance in the more numerous “other mesenchymal non-meningiothelial tumours” (a heterogeneous group, ranging in behaviour from lipomas to highly malignant sarcomas, that would not always be included in analyses of CNS tumours). For both of these categories, an unusually high proportion of patients resident in the low-volume regions were treated in the high-volume region, making the imbalance by treatment region even more marked and casting significant doubt on the assumed lack of bias.

3. The total number of patients diagnosed within life during the study period was 943, of whom 127 (13%) were excluded because of lack of histological confirmation. It would be of interest to know how these cases were distributed geographically and what were their survival rates. If certain tumours with an especially poor prognosis, such as mid-brain gliomas, were more likely to be biopsied, and hence included in the study series in a particular centre, then that centre’s overall survival rate would be biased downwards compared with other centres where such tumours were less likely to be biopsied. The description of the registry methods for updating diagnostic data also seems to imply that children whose diagnosis was not initially confirmed histologically, but for whom histology became available at a later operation, or even post mortem, might have been included.
4. While it was not the aim of this study to compare the Norwegian outcomes for various tumour types with the literature, such a comparative analysis is necessary in order to give the reader a framework within which to read the article. The only subgroup analysis to which we are given specific data relates to PNET/medulloblastoma. The 5-year overall survival rate for children living in and/or receiving treatment in the higher provider volume health region had an estimated 5-year overall survival rate of $42 \pm 7\%$ versus $65 \pm 6\%$ in the lower provider volume health region. On the SEER website [17] for the period 1995–2006 for children less than 15, the 5-year survival rate is 61.9%. Thus, the low-volume regions in Norway seem to be having results similar to those seen in the SEER database, whereas the results in the high-volume region would appear to be significantly different to that expected. While possible explanations for this include a failure to account for potential confounding factors or the impact of individual surgeon volume, it also may be

a genuine cause for specific concern. A further unexplained anomaly with this Norwegian data is the fact that there is no difference in 5-year survival in patients with supratentorial PNET compared with those “pure” medulloblastoma. In Fig. 3, the survival curves for the two geographically defined groups of patients only start to diverge around 1 year following diagnosis. This raises the question of whether at least some of the difference is attributable to variation between centres in the management of relapse.

5. Insufficient information is given regarding the “set-up” of paediatric neurosurgery in Norway. How is a paediatric neurosurgeon defined? Is a fellowship mandatory? How many surgeons are operating on paediatric tumours in each centre and what percentage of their workload is paediatric as opposed to adult? Is there a separate 24/7 paediatric neurosurgical service at each of the four departments? Who determines which neurosurgeon (adult or paediatric) operates on which tumours?

Conclusions

With many countries debating how best to deliver low-volume, high-cost, high-risk, specialist services, Solheim et al. [15] are to be congratulated on producing a timely article on provider volume and its relationship to outcome. The authors raise concerns over the use of centre volume as a proxy for quality, as they did not observe a relationship between centre volume and outcome in Norway. As this study raises more questions than it answers, an alternative conclusion from their study might be that it is not reasonable to wait another 20 years in order to have sufficient data to confirm that the Norwegian outcomes are comparable with other international centres. Only further centralisation would allow international comparative analysis to occur in a more timely fashion. Certainly, for this latter approach to be achieved it will be necessary for the international neurosurgical community to develop agreed outcome measures. It will also be necessary for healthcare providers to appreciate the importance of such data collection and to fund it appropriately.

Conflicts of interest None.

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