5(1), 1–10, 2023 | https://doi.org/10.1093/noajnl/vdad130 | Advance Access date 9 October 2023

Physician, patient, and caregiver support for a formal certification in pediatric neuro-oncology: A surveybased report from the SNO pediatrics working group

Holly B. Lindsay[®], Sylvia Cheng, Paul Graham Fisher, Katherine B. Peters[®], Kyle M. Walsh, David M. Ashley, and Annie Huang

Children's Hospital Colorado Center for Cancer and Blood Disorders, University of Colorado Anschutz Medical Campus, Aurora, Colorado, USA (H.B.L.); B.C. Children's Hospital, Division of Pediatric Hematology/Oncology/ BMT, Department of Pediatrics, The University of British Columbia, Vancouver, British Columbia, Canada (S.C.); Departments of Neurology and Pediatrics, Stanford University, Palo Alto, California, USA (P.G.F.); Preston Robert Tisch Brain Tumor Center, Department of Neurosurgery, Duke University School of Medicine, Durham, North Carolina, USA (K.B.P., K.M.W., D.M.A.); Division of Hematology Oncology, Department of Pediatrics, Hospital for Sick Children, University of Toronto, Toronto, Ontario, Canada (A.H.)

Corresponding Author: Holly B. Lindsay, MD, MS, Children's Hospital Colorado Center for Cancer and Blood Disorders, University of Colorado Anschutz Medical Campus, 13123 East 16th Avenue, Box B115, Aurora, CO 80045, USA (holly.lindsay@childrenscolorado.org)

Abstract

Background. Although CNS tumors are the most common pediatric cancer in the United States, most physicians caring for these patients are not formally certified in the subspecialty. To determine support for developing a formal certification process in pediatric neuro-oncology, the Society for Neuro-Oncology's Pediatrics Special InterestTrack Training and Credentialing working group performed a cross-sectional survey-based study of physicians and patients/caregivers of children with a CNS tumor history.

Methods. Surveys were built in Survey Monkey and were available for 3 months. The physician survey had 34 questions and was open to doctors currently caring for pediatric neuro-oncology patients. The patient/caregiver survey had 13 questions. Both surveys were completed anonymously.

Results. The physician survey was completed by 193 participants, the majority of whom self-identified as oncologists. Only 5.6% of survey participants had ever been board-certified in neuro-oncology; the majority of participating physicians were either unaware that this certification existed or thought they were not eligible due to training in pediatrics rather than neurology or internal medicine. Almost half of the self-identified pediatric neuro-oncologists had not completed any specific clinical neuro-oncology training. Over 75% of physicians were supportive of the implementation of a formal certification process in pediatric neuro-oncology. A total of 30 participants completed the patient/caregiver survey. Although the majority of survey participants were highly satisfied with their oncologist, 70% would have been more comfortable if their oncologist had been specifically certified in pediatric neuro-oncology. **Conclusions**. There is support from physicians, patients, and caregivers to establish a formal certification process in pediatric neuro-oncology.

Key Points

- >75% of surveyed physicians support a formal certification process for pediatric neuro-oncology.
- 70% of surveyed patients and caregivers support certification for pediatric neuro-oncologists.

© The Author(s) 2023. Published by Oxford University Press, the Society for Neuro-Oncology and the European Association of Neuro-Oncology.

This is an Open Access article distributed under the terms of the Creative Commons Attribution-NonCommercial License (https://creativecommons.org/licenses/by-nc/4.0/), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

Importance of the Study

Although CNS tumors are one of the most common diagnoses seen in pediatric oncology, very few pediatric neuro-oncologists are formally certified in this subspecialty. We identified that the majority of surveyed physicians were either not aware that certification in neuro-oncology existed at all or believed they were ineligible due to training in pediatrics rather than internal medicine or neurology. A significant majority of surveyed physicians, patients, and caregivers were supportive of the initiation of a formal certification process specifically in pediatric neuro-oncology. The components of this certification remain to be delineated by the pediatric neuro-oncology community, but we recommend starting by developing formal accreditation of pediatric neuro-oncology subspecialty fellowships.

CNS tumors represent 35% of all annual cancer diagnoses in U.S. patients ages 0-19 years old, accounting for more incident cases than either leukemias (28%) or lymphomas (17%).¹ CNS tumors are also the leading cause of cancerrelated death in children 0-14 years old in the United States,^{1,2} and pediatric CNS tumor incidence and mortality data are similar in population-based studies from other nations.³⁻⁷ Given the rapid pace of clinical and biologic discoveries in pediatric neuro-oncology as well as the complexity of caring for pediatric patients with brain and spinal cord tumors, the majority of moderate to large sized pediatric oncology programs have physicians that focus their practice either in part or in full on pediatric neurooncology. Pediatricians primarily in the United States and Canada may choose to undertake subspecialty training in pediatric neuro-oncology following completion of a fellowship in pediatric hematology-oncology or child neurology; however, this year of post-fellowship specialization is rare among U.S. clinicians caring for pediatric neurooncology patients. Recent data from the American Society of Pediatric Hematology/Oncology (ASPHO) shows that in 2020 <3% of pediatric hematology-oncology fellow graduates pursued additional formalizing training in pediatric neuro-oncology.8

In contrast, physicians providing neuro-oncology care for adult patients typically undergo a much more formalized training and certification process.⁹ After completion of a neurology residency or internal medicine residency followed by a medical oncology fellowship, most adult neuro-oncologists pursued further training in 1 of the 33 United Council for Neurological Subspecialties (UCNS)accredited neuro-oncology fellowship programs that are at least 12 months in duration, and most trained graduates subsequently sit for the 200 questions UCNS neurooncology certification examination. Of note, 25% of questions on this examination are focused on pediatric primary CNS tumors. Of the almost 300 physicians who are board-certified in neuro-oncology, less than 10 are pediatric neuro-oncologists.¹⁰

Given the rarity of pediatric brain tumors, there is a need to develop pediatric brain and spinal cord tumor Centers of Excellence across the globe to improve direct patient care, harmonize management strategies, and advance the field through collaborative clinical research studies. Consequently, key members of the Society for Neuro-Oncology (SNO) leadership team as well as the members of the SNO pediatrics special interest track training and credentialing working group believe it would be beneficial for physicians caring for pediatric neuro-oncology patients to train in an accredited subspecialty pediatric-specific neuro-oncology fellowship program and subsequently to undergo a separate and specialized formal certification process similar to what is done by adult neuro-oncologists. To determine if our belief was generalizable, we surveyed both physicians actively caring for patients with pediatric CNS tumors and patients or caregivers of children with a history of pediatric CNS tumors to determine their opinions on formalizing post-fellowship training for pediatric neuro-oncologists.

Materials and Methods

The pediatric special interest track training and credentialing working group was formed by SNO and began meeting monthly in May 2021. Our observational study collected data via 2 cross-sectional surveys generated by the working group: 1 for physicians caring for pediatric patients with CNS tumors and 1 for patients with a history of a CNS tumor or their caregiver. Surveys were built in Survey Monkey and generated in English only for physicians and in English, Spanish, and French for patients/ caregivers. Prior to releasing the surveys for completion, the research project was approved by the Baylor College of Medicine Institutional Review Board (IRB). A waiver of the requirement for written documentation of informed consent was granted by the IRB as completion of the survey was considered consent for this research. Surveys were live and able to accept responses for a total of 3 months.

Physician Survey

The primary objective of the physician survey was to assess the perspectives and opinions of practicing pediatric oncologists and/or pediatric neuro-oncologists regarding the role of a formal training and certification process for pediatric neuro-oncologists. The secondary objectives were to: (1) Describe the demographics and scope of practice of current pediatric oncologists including characterizing the patient population cared for; (2) Quantify the number of current or previously board-certified pediatric neuro-oncologists; (3) Quantify physician-identified gaps in pediatric neuro-oncology training, and (4) Identify which was a PhD earned by 51 participants.

workplace resources at institutions relevant to care of peguantify the survey response rate. A total of 48.9% (92/188) diatric neuro-oncology patients. The physician survey was of participants were medically fluent in 1 of 34 languages emailed to a list serve of physicians who had previously atin addition to English; the most common languages were tended an SNO-sponsored conference and to members of Spanish (15.4%, 29/188), French (10.6%, 20/188), and Hindi both the International Society of Paediatric Oncology and (5.3%, 10/188). A total of 37.2% of survey participants the Canadian C17 Children's Cancer and Blood Disorders; (71/191) lived in 1 of 37 countries other than Canada or the the survey was also posted in the member forum of the USA. Survey participants held a total of 96 advanced de-ASPHO and on the SNO Twitter feed. The survey including grees in addition to MD/MBBS/DO, with most common of

Scope of practice.—The majority of survey participants (71.7%, 129/180) identified as oncologists, while 11.1% (20/180) were neurologists. Only 5.6% of participants (10/180) had ever been board-certified in neuro-oncology through the UCNS. Almost 70% of survey participants (69.9%, 114/163) did not obtain UCNS neuro-oncology board certification either because they were unaware of the certification or thought they were ineligible. Another 10% (10.5%, 17/163) did not feel the certification was meaningful to their career or patients. Of note, physicians are eligible to obtain UCNS neuro-oncology board certification in neurology, child neurology,

Patient/Caregiver Survey

neuro-oncology.

The primary objective of the patient/caregiver survey was to assess the relationship of patients treated for a pediatric CNS tumor (or their caregiver) with their oncologist. The secondary objective was to determine support from this population for developing and recommending a formal training and certification process for pediatric neuro-oncologists. Invitations to complete the survey were emailed to list serves from the following organizations: (1) Alex's Lemonade Stand Foundation, (2) American Brain Tumor Association, (3) Children's Brain Tumor Foundation, (4) Michael Mosier Defeat DIPG Foundation, (5) National BrainTumor Society, (6) Pediatric Brain Tumor Foundation, and (7) Rainbows and Smiles. The survey including the introductory letter is shown in Supplementary Figure 2.

the introductory letter is shown in Supplementary Figure 1.

no participant would respond to all questions as some

were automatically skipped based on responses to prior

questions. Any physician currently caring for pediatric

patients (≤21 years of age) with CNS tumors was eligible

for survey participation; nonphysician providers were

excluded. Participants took the survey anonymously,

though they were given the option to provide their

name and email address. Survey topics included dem-

ographics, scope of practice, self-assessment of gaps in

pediatric neuro-oncology training, workplace resources,

and opinions regarding formal certification in pediatric

The survey included a total of 34 questions, though

The survey included up to 13 questions and was completed anonymously. Survey topics included information about the respondents' tumor diagnosis (including age at diagnosis, tumor pathology, and tumor location), treatment (including being offered enrollment on a clinical trial), and treating oncologist (including if they mainly saw pediatric or adult patients and their willingness to facilitate the patient receiving a second opinion), as well as their opinion of a formal pediatric neuro-oncology training and certification process for their oncologist.

Results

Physician Survey

Demographics.—A total of 193 participants completed the survey; their demographics are shown in Table 1 and Figure 1. Due to the numerous ways this survey could be accessed by physicians (ie, via email invitation, clicking survey link from the Twitter post, etc.), we were unable to

Table 1. Demographics of Physician Survey Participants

| N = 193 respondents | Number of re- sponses | % of respondents with response | | | |
|--|--------------------------|--------------------------------|--|--|--|
| Gender* | | | | | |
| Female | 91 | 47.4% | | | |
| Male | 97 | 50.5% | | | |
| Non-binary | 1 | 0.5% | | | |
| Prefer not to say | 3 | 1.6% | | | |
| Age | | | | | |
| <30 years | 1 | 0.5% | | | |
| 30–50 years | 116 | 60.1% | | | |
| 50–54 years | 20 | 10.4% | | | |
| 55–65 | 43 | 22.3% | | | |
| >65 years | 12 | 6.2% | | | |
| Prefer not to say | 1 | 0.5% | | | |
| Years since completion of final clinical training* | | | | | |
| <1 year | 11 | 6.0% | | | |
| 1–5 years | 29 | 15.9% | | | |
| 6–10 years | 33 | 18.1% | | | |
| 11–15 years | 43 | 23.6% | | | |
| 16–20 years | 23 | 12.6% | | | |
| >20 years | 43 | 23.6% | | | |
| Percent of work time spent clinically (including both patient care and clinical research)* | | | | | |
| 90%-100% | 62 | 34.1% | | | |
| 75%–89% | 52 | 28.6% | | | |
| 50%–74% | 37 | 20.3% | | | |
| 25%–49% | 16 | 8.8% | | | |
| <25% | 15 | 8.2% | | | |
| <i>Note</i> : *All respondents did not respond to these questions. | | | | | |



neurological surgery, internal medicine and medical oncology, pediatrics and pediatric hematology-oncology, or radiation oncology from an American Board of Medical Specialties (ABMS) certifying board, equivalent certification from the Royal College of Physicians and Surgeons of Canada or the American Osteopathic Association, or an appropriate board of the European Union of Medical Specialties.¹¹ For applicants applying under the "internationally trained faculty pathway" that are certified in their primary specialty by a competent medical board of their country of origin, this board must be approved by the UCNS Certification Council prior to the applicant being deemed eligible to obtain UCNS neuro-oncology certification.

A total of 77.2% of participants (132/171) completed a residency in pediatrics or medicine-pediatrics and were eligible for completion of the remainder of the survey, though only 124 participants continued participation after this question. Over 80% (84.7%, 105/124) of the remaining 124 survey participants saw only or mostly pediatric patients in their practice rather than adult oncology patients >22 at diagnosis (Figure 1A). Over half of the survey

participants primarily considered themselves pediatric neuro-oncologists (60.5%, 75/124) rather than general pediatric oncologists, hematologist-oncologists, or neurologists (Figure 1B). However, of these almost half (41.9%, 52 responses from 124 respondents) did not complete any specific clinical neuro-oncology training either during or after their pediatric hematology-oncology or child neurology fellowship. A pediatric neuro-oncology fellowship was not offered by over ²/₃ of institutions (70.7%, 87/123) where participants completed their fellowships.

Self-assessment of gaps in pediatric neuro-oncology training.—Over 50% of participants (52.0%, 64/123) spent at least 20% of their clinical time during pediatric hematology-oncology or child neurology fellowship caring for patients with CNS tumors. Survey participants had a wide variety of comfort levels in the care of pediatric patients with CNS tumors (Table 2). It was relatively rare for participants to refer pediatric patients with CNS tumors to other institutions other than for consideration of a clinical trial. There was a wide variety of areas within pediatric

| N = 124 respondents | Extremely com- fortable | Very comfort- able | Somewhat com- fortable | Not comfort- able |
|---|----------------------------|-----------------------|---------------------------|----------------------|
| Creating a treatment plan for newly diagnosed tumors | 58.9% (73/124) | 25.8% (32/124) | 11.3% (14/124) | 4.0% (5/124) |
| Creating a treatment plan for recurrent/progressive tumors* | 41.5% (51/123) | 33.3% (41/123) | 17.1% (21/123) | 8.1% (10/123) |
| Managing chemotherapy side effects | 75.8% (94/124) | 20.2% (25/124) | 2.4% (3/124) | 1.6% (2/124) |
| Managing patients enrolled on clinical trials* | 66.7% (82/123) | 22.8% (28/123) | 7.3% (9/123) | 3.3% (4/123) |
| Managing radiation side effects | 39.5% (49/124) | 44.4% (55/124) | 14.5% (18/124) | 1.6% (2/124) |
| Utilizing high dose chemotherapy with autologous stem cell rescue | 47.6% (59/124) | 25.8% (32/124) | 14.5% (18/124) | 12.1% (15/124) |
| Managing palliation and end of life care | 47.6% (59/124) | 40.3% (50/124) | 12.1% (15/124) | 0.0% (0/124) |
| Note: | | | | |

*1 respondent did not respond to these questions.

 Table 3.
 Areas of Additional Training in Pediatric Neuro-oncology Physician Survey Participants Felt they would have Benefitted from Prior to

 Starting Independent Practice (participants could choose more than 1 response and could enter free text entries)

| | Number of responses | % of respondents with response |
|--|---------------------|--------------------------------|
| | Number of responses | % of respondents with response |
| Survey-provided response options | | |
| More time spent making treatment plans/ decisions for pediatric patients with CNS tumors | 54 | 47.0% |
| Dedicated time with pediatric neuro- radiologist | 52 | 45.2% |
| Seeing more pediatric patients with CNS tumors | 47 | 40.9% |
| Seeing more pediatric patients specifically with rare CNS tumors | 44 | 38.3% |
| Seeing more pediatric patients specifically with recurrent CNS tumors | 44 | 38.3% |
| Dedicated time with Radiation Oncologist | 39 | 33.9% |
| More time caring for patients enrolled on clinical trials | 35 | 30.4% |
| Other (no free text entry included by partic- ipant) | 11 | 9.6% |
| Free texted entry | | |
| Dedicated time with pediatric neurologist | 1 | 0.9% |
| | Total respondents: | 115 |
| | | |

neuro-oncology which survey participants felt they would have benefitted from prior to starting independent practice; the most common areas were (1) More time spent making treatment plans/decisions for pediatric patients with CNS tumors (47.0%, 54 responses from 115 respondents) and (2) Dedicated time with a pediatric neuro-radiologist (45.2%, 52 responses from 115 respondents; Table 3).

Workplace resources.—Over 80% of our participants (83.7%, 133/159) practiced at an academic center. As expected based on those data, the majority of survey participants had access at their institution to resources integral to the care of pediatric patients with CNS tumors, including pediatric neurologists, neurosurgeons,

neuro-psychologists, neuro-ophthalmologists, and radiation oncologists as well as the capacities for harvesting autologous stem cells and performing molecular tumor analyses.

Opinion regarding formal certification in pediatric neuro-oncology.—Over 75% of participants were either "somewhat supportive" (32.1%, 51/159) or "very supportive" (47.8%, 76/159) of the implementation of a formal certification process specifically for pediatric neuro-oncology, while just over 10% were "not at all supportive" (4.4%, 7/159) or "not very supportive" (5.7%, 9/159; Figure 1E). Responses were mixed regarding what should be included in the formal certification process, Table 4. Reasons Physician Survey Participants were Unlikely to take a Certifying Exam in Pediatric Neuro-oncology if Available (participants could enter free text entries)

| | Number of responses | % of respondents with response |
|--|---------------------|--|
| | Number of responses | ⁷⁶ of respondents with response |
| Survey-provided response options | | |
| l do not feel that formal board certification in pediatric neuro-oncology would be beneficial to my career | 21 | 36.8% |
| l do not feel that formal board certification in pediatric neuro-oncology is meaningful to my patients/families | 13 | 22.8% |
| Free texted entries | | |
| I am already too senior in my career | 7 | 12.3% |
| No time to study | 4 | 7.0% |
| Most of my clinical practice is not in pediatric neuro- oncology | 3 | 5.3% |
| Not interested in taking the test | 3 | 5.3% |
| I do not live in the US | 2 | 3.5% |
| I am already certified in adult neuro-oncology | 1 | 1.8% |
| l did not do a formal fellowship so don't think I would be eligible for the exam | 1 | 1.8% |
| Most of my practice is adult patients | 1 | 1.8% |
| Other (no free text entry included by participant) | 1 | 1.8% |
| | Total respondents: | 57 |
| | | |

with only 40% (40.9%, 63/154 respondents) agreeing that a written examination should be included. However, despite these generally positive responses toward the idea of formalizing certification in pediatric neuro-oncology, just over 40% of survey participants (40.1%, 63/157) were "not likely" to take the certifying examination if a formal board certification examination was offered; only 32.5% (51/157) responded they were "very likely" to take the examination (Figure 1F). Reasons for not being likely to take the examination varied but most commonly were feeling that the board certification would not be beneficial to the participants' careers (36.8%, 21/57) or to the patients/families they treat (22.8%, 13/57; Table 4).

Patient/caregiver survey.—A total of 30 participants (either the patient themselves or their caregiver) completed the English version of the survey; no responses were submitted for either the Spanish or French versions (Figure 2). As the list serves of the 7 organizations to whom survey invitations were sent were not shared with the authors, we are unable to quantify the response rate to the survey. There was a wide age distribution at CNS tumor diagnosis, with the most common age range being 6-9 years (27.6%, 8/29) (Figure 2A). The most common tumor was low-grade glioma (8/30, 26.7%) followed by medulloblastoma and germ cell tumor (both 13.3%, both 4/30) (Figure 2A). All 28 survey participants who remembered the primary location of their tumor had intracranial rather than spine disease. Most participants received therapy in the United States. Almost ¹/₃ of survey participants (31.0%, 9/29) were cared for by an oncologist who saw "mainly" or "mostly" adult patients. Over 1/4 of participants (26.7%, 8/30) lived at least 60 miles from the hospital during their treatment. Over half of the participants received neurosurgery (73.3% of respondents, 22/30), chemotherapy (63.3% of respondents, 19/30), and/or radiation therapy (56.7% of respondents, 17/30) as a component of their anti-neoplastic therapy (Figure 2C). Almost half of the survey participants (45.5%, 10/22) received treatment on a clinical trial at some point during their therapy while another 13.6% of participants (3/22) were considered for enrollment in a study but did not qualify (Figure 2D).

Over half of the survey participants (55.2%, 16/29) discussed the option of getting a second opinion with their oncologist. Over 3/4 of participants (81.3%, 13/16) felt that this conversation went "well." Of the survey participants who did not discuss the option of getting a second opinion with their oncologist, only 1 participant (8.3%, 1/12) was "scared that my doctor would be hurt or offended by this request." Overall, on a scale of 1 to 9, where 1 was "I was not at all satisfied with the care I received from my doctor" and 9 was "I felt that my doctor was an expert in my tumor and provided excellent care," the vast majority of survey participants (85.7%, 18/21) scored their satisfaction with their oncologist at 7 or higher (Figure 2E). However, 70% (14/20) reported that they would have been more comfortable if their oncologist had been specifically certified in pediatric neuro-oncology (Figure 2F).

Discussion

This survey-based study demonstrates that there is support both from physicians caring for pediatric patients with CNS tumor and patients/caregivers with CNS tumors for the establishment of a formal certification process in



pediatric neuro-oncology. The challenge is to determine how this can best be achieved to meet the goal of optimizing the care of pediatric neuro-oncology patients while not disincentivizing trainees to pursue careers in this field through additional training and certification. When interpreting our data, it is important to note that 62.8% of physician participants were from the United States and Canada, which may not be representative of the global landscape of subspecialist training. We additionally acknowledge that our survey results are limited to reflect the opinions only of those physicians and patients/caregivers who chose to respond; additionally, due to the multiple means by which the physician and patient/caregiver survey could be accessed, we are unable to assess any possible contribution from response bias to our data.

We recommend that the first step in formalizing a certification in pediatric neuro-oncology is to establish accreditation to centers offering pediatric neuro-oncology fellowships. This accreditation could be through the ACGME, UCNS, or a separate pediatric neuro-oncology specific group which would need to be established from scratch. Accreditation will allow standardization of an educational curriculum with the goal of ensuring consistency and quality amongst fellowship programs. Regardless of the accrediting body, we propose that to be eligible for accreditation, pediatric neuro-oncology programs must be at least 1 year in duration following pediatric hematologyoncology or child neurology fellowship and include (1) Dedicated time with pediatric neuro-pathologists and molecular pathologists, pediatric neuro-radiologists, pediatric palliative care physicians, and radiation oncologists, (2) Treatment of patients with modalities including chemotherapy, high dose chemotherapy followed by autologous stem cell rescue, molecularly-targeted agents, neurosurgery, and radiation therapy, (3) Diagnosis and management of pediatric patients with both newly diagnosed and recurrent/progressive/refractory CNS malignancies including the opportunity to provide second opinions, (4) Care of patients in both inpatient and outpatient settings including the critical care unit and long-term survivor/late effects clinic, (5) Determining eligibility for and enrolling patients on clinical trials, (6) Managing patients with germline alterations predisposing to CNS malignancies, and (7) Active participation in (and ideally leadership

Neuro-Oncology Advances 8

of) a recurring multi-disciplinary pediatric neuro-oncology tumor board. Additionally, as pediatric CNS diagnoses are increasingly recognized to be highly clinically and biologically heterogenous, we would suggest that a minimum number of patients should be established for a variety of CNS tumor diagnoses that trainees must see in order to graduate from an accredited pediatric neuro-oncology subspecialty fellowship.

An important consideration in the formalization of pediatric neuro-oncology training is the significant difference in exposure to patients with CNS tumors seen in a pediatric hematology-oncology fellowship compared with a child neurology fellowship. Care for patients with solid tumors of the central nervous system is a core competency of the program requirements for graduate medical education in pediatric hematology-oncology as defined by the Accreditation Council for Graduate Medical Education (ACGME).12 17% of questions on the American Board of Pediatrics (ABP) subspeciality initial certification examination in pediatric hematology-oncology is focused on solid tumors, which includes guestions on medulloblastoma, low- and high-grade gliomas, ependymoma, CNS germ cell tumors, and rare brain tumors.¹³ In contrast, the ACGME requirements for graduate medical education in child neurology require only 12 months of training in clinical child neurology, with a statement that neuro-oncology "should" be included in the teaching curriculum.¹⁴ Furthermore, of the 50% of questions on the American Board of Psychiatry and Neurology, Inc. certification examination in neurology with special qualification in child neurology focused on neurologic disorders and topics, only 1%–3% are related to neuro-oncology.¹⁵These significant discrepancies in focus on pediatric neuro-oncology during pediatric hematology-oncology and child neurology fellowship especially highlight the need for further subspecialty training focused on CNS tumors for child neurologists interested in a career as a pediatric neuro-oncologist.

As we look to creating a formalized pediatric-specific neuro-oncology certification, it is important to note that assessment in medical education has been moving away from traditional learning models that focus on memorization and content mastery toward a competency-based approach whereby learning is adapted to achieve the required skills, knowledge, qualification, or capacity.¹⁶ It is consequently important to incorporate both knowledge and experience in pediatric neuro-oncology education and program structures and ensure assessment measures truly reflect achievement in needed competencies and skills. Clearly, then, the question of whether a board examination taken after completion of an accredited subspecialty fellowship should be required for pediatric neuro-oncology certification is complex. Although almost half of physician survey respondents were "very supportive" of implementing a formal certification process in pediatric neuro-oncology and another 1/3 of participants were "somewhat supportive," there was overall much less agreement about what should be included/required as part of the certification process; in fact, less than half of physician survey respondents (40.9%, 63/154) felt that a written examination should be included as part of the formal certification process.

Although fellowship-trained pediatricians who train in a UCNS-accredited fellowship program or are deemed appropriate via the practice track certification option are currently eligible to become certified in neuro-oncology by the UCNS, the overwhelming majority of self-identified pediatric neuro-oncologists have not completed this accreditation mainly attributed to not being aware or not knowing they were eligible to write a certification examination. Our data also highlight the likely misalignment between important competencies and skills needed by pediatric neurooncologists and what is currently being assessed by the majority of the UCNS neuro-oncology board examination, which skews heavily toward adult neuro-oncology. As currently written, the UCNS neuro-oncology certification examination includes 4 content areas, somewhere around 50% is applicable to pediatric-trained physicians, as 25% of the questions are focused on pediatric primary CNS tumors and another 25% covers systemic cancer-related neurologic disorders and side effects of anti-neoplastic therapies; the remaining 50% of questions cover adult primary CNS tumors and metastatic cancer to the nervous system (rarely seen in pediatric oncology and if present managed by pediatric oncologists focused on the primary tumor type rather than a neuro-oncologist).

Assuming that a new pediatric-focused certification examination is created, this testing could be run through several organizations including the ABP, UCNS, or a separate pediatric neuro-oncology specific group which would need to be established from scratch. We find the idea of an oral (rather than written) certifying examination including standardized patient cases to be intriguing, as it assesses not only candidates' knowledge but also diagnostic and communication skills. Although no oral assessment is currently part of any ABP certification, 14 of the 24 ABMS specialty boards include oral certifying examinations.¹⁷ It would be important to offer the oral examination both in-person at various locations as well as virtually to increase the number of candidates that can participate.¹⁸ Ideally, a formal certification in pediatric neuro-oncology would be open to international trainees who seek to provide more expert care in their home countries, outside the United States and Canada. Of note, as of the publication of this article, the authors and members of the SNO pediatrics special interest track training and credentialing working group have not had any formal discussions with the leadership of the ABP, ACGME, or UCNS regarding the establishment of pediatric neuro-oncology fellowship accreditation or creation of a board certification. This publication was designed to further engage and stimulate discussions related to these topics within the pediatric neuro-oncology physician community.

This article does not presume to recommend that other "sub"-subspecialties within pediatric hematologyoncology (ie, hematopoietic stem cell transplantation and cellular therapies, sarcomas, hemostasis, and thrombosis) should have a recommended or required additional year of clinical training and board certification, as our investigation focused only on pediatric neuro-oncology. However, many physicians with a dedicated clinical focus in a specific niche within pediatric hematology–oncology (rather than working as a "generalist") have undertaken additional post-fellowship training within their area of expertise. The topic of formalizing "sub"-subspecialty training and certification in pediatric hematology–oncology is being addressed by fields outside of neuro-oncology, as represented by a 2018 survey-based publication undertaken to assess support for a dedicated ACGME-accredited training program and certification examination in the subspecialty of adult and pediatric bone marrow transplantation (BMT).¹⁹ Unlike our results, however, the majority of the BMT physician community did not support formalizing certification in this field due to it being "burdensome"; however, over half did feel that certification in BMT would "improve education."

Importantly, if a formal certification process is adopted in this field, it will be necessary to monitor the impact on patient care, including access and both health-related quality of life and survival outcomes, despite challenges in quantifying these metrics. If there is a decision by the pediatric neuro-oncology community to begin centralizing care of these patients under pediatric neuro-oncology accredited fellowship trained and "sub"-subspecialty board-certified doctors, it will be key to ensure that patients diagnosed in rural communities or smaller institutions have access to trained providers via telemedicine consultation with or co-management by experts at academic centers. The option for remote management by trained pediatric neuro-oncologists with the active treatment provided by local general oncologists would allow all patients to receive treatment plans from disease experts; certainly, patients located in rural communities or cared for at smaller centers without a dedicated pediatric neurooncologist would benefit from traveling to receive specific interventions such as radiation therapy, neurosurgery, and/ or autologous hematopoietic stem cell transplantation at an academic center, but could receive numerous other interventions nearer their homes such as standard-dose chemotherapy, transfusions, and treatment toxicity monitoring. Currently, in the United States, there are insurance agreements in place for less populated states to allow patients to be seen and treated at out-of-state expert centers. Most of these regional referral centers also budget to allow various faculty to obtain out-of-state medical licenses for nearby states to facilitate telemedicine visits and strategically plan oncology and pathology physician staffing to take into account not just local but regional patients. These centers also typically work closely with social work colleagues to provide housing, transportation, and financial assistance for patients and families coming from noncommutable distances. We certainly acknowledge the challenge this would be for patients from rural areas but feel that all steps should be taken to ensure that they are able to receive expert-level care just like those located near a trained pediatric neuro-oncologist.

Additionally, it will be important to monitor if the number of trainees and attending physician workforce can support this limitation in those caring for pediatric patients with CNS tumors. Almost 40% of the physician survey participants did not consider themselves pediatric neuro-oncologists despite caring for this patient population; it is possible that limiting or prioritizing the care of these patients to "sub"subspecialty-trained physicians might upset doctors that identify as general hematologists-oncologists and/or lead to the perception that the provider options for this population are restrictively limited. We also acknowledge that not all trainees may have the opportunity to pursue specializing training in pediatric neuro-oncology due among other reasons to financial reasons (continuing to receive a trainee salary rather than faculty salary) or location. However, of the 84 accredited pediatric hematology-oncology in Canada and the United States, almost 1/3 of programs (25/84, 29.8%) offer pediatric neuro-oncology sub-specialty fellowships programs, representing 3 provinces, 17 states, and the District of Columbia.²⁰ We further acknowledge that the cost of an additional board examination may be costprohibitive for some physicians, especially in the setting of the additional costs of maintaining their specialty and subspecialty certifications. Finally, it is possible that additional training with certification could deter some candidates from pursuing a lifelong clinical career or niche in brain tumors; however, we are unaware of any accreditation or certification process in any subspeciality to date that has seen that consequence.

Given the wide variety of CNS tumor pathologies, complexity of care for these diagnoses, and rapid pace of biologic discoveries being quickly translated into clinical care, we strongly feel that this change will optimize the care of pediatric neuro-oncology patients by allowing their treatment plan to be created and managed by disease experts. We want to highlight however that our recommendation is not to mandate that every physician caring for pediatric patients with CNS tumors be trained in an accredited pediatric neuro-oncology fellowship and board-certified in the field. We instead are recommending that fellowship accreditation and board certification in pediatric neuro-oncology be established and formalized so that there are true disease experts available to care for this unique patient population.

In summary, there is support from both physicians caring for pediatric patients with CNS tumors and the patients and their caregivers to establish a formal training and certification process in pediatric neuro-oncology. Although the details of this certification remain to be established, we feel strongly that this will allow children with brain and spinal cord tumors to receive oncologic care from the most optimally trained physicians and will facilitate internationally harmonized clinical standards of practice, leading to critical advancements in our clinical and biological investigations into CNS tumors.

Supplementary material

Supplementary material is available online at *Neuro-Oncology* (https://academic.oup.com/neuro-oncology).

Keywords:

accreditation | certification | pediatric neuro-oncology | training

Funding

This work was not supported by any funding sources.

Acknowledgments

We acknowledge the efforts of International Society of Paediatric Oncology Chief Executive Officer Susanne Wollaert and C17 Council Executive Director Kathy Brodeur-Robb for help disseminating the physician survey and both Mark Mosier, JD, and Nancy Goodman, JD for their help designing and disseminating the patient/caregiver survey. We appreciate the support of SNO leadership including the Executive Director J. Charles (Chas) Haynes, JD, and the Director of Administration Shelley Pressley, MBA, CMP.

Conflicts of interest statement

The Society for Neuro-Oncology supported this study and assisted with survey dissemination. There is no other conflict of interests from any author related to this study.

Authorship statement

Study design: H.B.L., S.C., P.G.F., K.B.P., K.M.W., D.M.A., A.H.; Survey creation: H.B.L., S.C., P.G.F., K.B.P., K.M.W., D.M.A., A.H.; Survey dissemination: H.B.L., S.C., P.G.F.; Data analysis and interpretation: H.B.L., S.C.; Writing of the manuscript: H.B.L., S.C., P.G.F., K.B.P., K.M.W., D.M.A., A.H.; Approval of final version: H.B.L., S.C., P.G.F., K.B.P., K.M.W., D.M.A., A.H.

References

- Ostrom QT, Price M, Ryan K, et al. CBTRUS statistical report: pediatric brain tumor foundation childhood and adolescent primary brain and other central nervous system tumors diagnosed in the United States in 2014-2018. *Neuro Oncol.* 2022;24(Suppl 3):iii1–iii38.
- (CBTRUS) CBTRotUS. Fact Sheet for Pediatric Brain Tumors. https:// www.soc-neuro-onc.org/UploadedFiles/CBTRUS%20Fact%20 Sheet%20for%20Pediatric%20Brain%20Tumors.pdf. 2022.
- Cheng S, McLaughlin JR, Brown MC, et al. Maternal and childhood medical history and the risk of childhood brain tumours: A case-control study in Ontario, Canada. *Br J Cancer*. 2023;129(2):318–324.

- Steliarova-Foucher E, Colombet M, Ries LAG, et al.; IICC-3 contributors. International incidence of childhood cancer, 2001-10: A populationbased registry study. *Lancet Oncol.* 2017;18(6):719–731.
- Youlden DR, Baade PD, Green AC, et al. The incidence of childhood cancer in Australia, 1983-2015, and projections to 2035. *Med J Aust.* 2020;212(3):113–120.
- Youlden DR, Henshaw C, Gottardo NG, Hassall T, Aitken JF. Incidence and survival for childhood central nervous system tumours in Australia, 1983-2016. *J Neurooncol*. 2021;155(2):203–213.
- Wellbrock M, Spix C, Ronckers CM, et al. Temporal patterns of childhood cancer survival 1991 to 2016: A nationwide register-study based on data from the German Childhood Cancer Registry. *Int J Cancer*. 2023;153(4):742–755.
- Hastings C, Borinstein SC, Bergsagel DJ, et al.; American Society of Pediatric Hematology Oncology Workforce Publication Workgroup. The American Society of Pediatric Hematology Oncology workforce, productivity, and fellowship assessment: Current state of the workforce. *Pediatr Blood Cancer.* 2023;70(5):e30221.
- Gonzalez Castro LN, Forst DA, Plotkin SR, Lee EQ. A practical guide to neuro-oncology fellowship. J Neurooncol. 2022;156(1):73–79.
- Peters KB, Patel M, Mrugala M. QLTI-25 creating a chemotherapy competency metric for neuro-oncology fellowship trainees. *Neuro-Oncology*. 2022;24(Suppl 7):vii240–vii240.
- Araki A, Chocholous M, Gojo J, et al. Chromosome 1q gain and tenascin-C expression are candidate markers to define different risk groups in pediatric posterior fossa ependymoma. *Acta Neuropathol Commun.* 2016;4(1):88.
- 12. https://www.acgme.org/globalassets/PFAssets/ ProgramRequirements/327_PediatricHematologyOncology_2020. pdf?ver=2020-06-29-162712-443&ver=2020-06-29-162712-443.
- 13. https://www.abp.org/sites/abp/files/outline-hemo.pdf.
- 14. https://www.acgme.org/globalassets/pfassets/ programrequirements/185_childneurology_2023.pdf.
- https://www.abpn.com/wp-content/uploads/2020/11/2021_Child_ Neurology_CERT_Content_Specifications.pdf.
- Fraser AB, Stodel EJ, Jee R, Dubois DA, Chaput AJ. Preparing anesthesiology faculty for competency-based medical education. *Can J Anaesth.* 2016;63(12):1364–1373.
- Mehta A, Allen B, Goldman JJ. Oral boards: dissecting the differences among medical specialties. *Plast Reconstr Surg Glob Open*. 2023;11(1):e4729.
- Furnas HJ. Overcoming barriers to board certification. JAMA Surg. 2023;158(8):785.
- Malone MRL AK, Perales M, Costa LJ, et al. 371 Should BMT become a certified subspecialty? Results of the 2017 Board Certification Survey. *Biol Blood Marrow Transplant*. 2018;24(Suppl 3):S305–S306.
- 20. https://www.soc-neuro-onc.org/WEB/Resources/ Pediatric_Fellowship_Resources/WEB/Resources_Content/ Pediatric_Neuro-Oncology_Fellowship_Programs. aspx?hkey=2f8b4fb7-e639-4c16-8a45-6eb25c87f1bf.