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Pediatric neurosurgical bellwether procedures for infrastructure capacity building in hospitals and healthcare systems worldwide

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Abstract

Purpose Quantifying the global burden of pediatric neurosurgical disease—and current efforts addressing it—is challenging, particularly in the absence of uniform terminology. We sought to establish bellwether procedures for pediatric neurosurgery, in order to standardize terminology, establish priorities, and facilitate goal-oriented capacity building.

Methods Members of international pediatric neurosurgical and pediatric surgical societies were surveyed via the Research Electronic Data Capture (REDCap) platform. Among 15 proposed neurosurgical procedures, respondents assigned numerical grades of surgical necessity and selected hospital-level designation within a three-tiered system. A procedure was considered a bellwether if (a) the majority of respondents deemed it necessary for either a primary- or secondary-level hospital and (b) the procedure was graded at or above the 90th percentile on a continuous scale of essentiality. Data were compiled and analyzed using Stata software.

Results Complete responses were obtained from 459 surgeons from 76 countries, the majority of whom practiced in a tertiary referral hospital (88%), with a primarily public patient population (64%). Six bellwether procedures were identified for pediatric neurosurgery: shunt for hydrocephalus, myelomeningocele closure, burr holes, trauma craniotomy, external ventricular drain (EVD) insertion, and cerebral abscess evacuation. Few differences in bellwether criteria designations were observed among respondents from different World Health Organization regions and World Bank income groups.

Conclusions The six bellwether procedures identified can be used as markers of infrastructure capacity at various hospital levels, hence allowing targeted neurosurgical capacity-building in low-resource settings in order to avert disability and death from childhood neurosurgical disease.

Keywords Bellwether · Global · Essential surgery · Pediatric neurosurgery · Surgery · Worldwide

Introduction

Each year, nearly 23 million people worldwide are estimated to suffer from a neurological insult or injury that requires the expertise of a neurosurgeon [1]. The vast majority of this disease burden emanates from low- and middle-income countries (LIC/MICs) where surgical resources are limited [2]. The proportion of neurological disease burden specifically affecting children is unknown, but expected to be substantial given that children comprise a greater proportion of the population in LIC/MICs relative to high-income countries (HICs) [3]. Pediatric surgical disease is an oft-neglected component of the healthcare sector in many resource-poor countries, where surgical systems are expensive to establish

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and difficult to maintain [4, 5]. Settings with inadequate surgical coverage for children can be challenging to identify for a myriad of reasons: sparse casualty department records, inconsistent hospital surgical logs, overlapping but non-collaborative platforms for healthcare delivery, and pre-hospital disease burden and mortality.

The term "bellwether procedure" was coined by O'Neill and colleagues to describe a fundamental surgical procedure that, when recorded and studied, could facilitate the assessment of a hospital's ability to perform essential surgical care [6]. A bellwether procedure is itself an essential surgical procedure and the rate, safety, and efficacy at which it is performed is an indication of a given healthcare system's capability of providing adequate surgical services to the at-risk population. Among the numerous surgical procedures that fall within the armamentarium of a surgical specialist, defining which procedures are of bellwether status is important for three primary reasons. First, it establishes priorities among community healthcare workers and regional hospitals by drawing attention to otherwise neglected pathologies. Second, it facilitates and simplifies data collection and serves as a metric by which to monitor the activity of surgical departments [6]. Lastly, the designation of bellwether procedures carries implications for governmental and external aid organizational prioritization and funding. Bellwethers for pediatric neurosurgery have not been established. In this global survey of pediatric surgical care providers, essential procedure valuation and hospital-level designations are merged to propose a defined set of bellwether procedures for the field of pediatric neurosurgery.

Methods

An initial invitation email was sent July 9th of 2017 to members of the International Society for Pediatric Neurosurgery (ISPN), the European Society for Pediatric Neurosurgery (ESPN), the Global Initiative for Children's Surgery (GICS), and the World Federation of Associations of Pediatric Surgeons (WOFAPS). The email contained a link to an approximately 15-min survey housed within the REDCap (Research Electronic Data Capture) data manager at Vanderbilt University [7]. Two reminder emails were sent, and answers were collected until August 20th of 2017.

Candidate neurosurgical procedures were informed by anecdotal case volume data and author consensus. Initially, broad surgical procedures or disciplines were presented; respondents were asked to select whether the given procedures were commonly performed at their institution. It was left to the respondent to determine with what frequency a procedure was performed to be deemed common. Next, the broad categories were subdivided into more specific entities; 15 procedures were presented to survey respondents for consideration as bellwether procedures (Table 2).

Answers were exported directly to Stata® software version 14 for analysis. Answers were reported as counts, and relative proportions were labeled as percentages. Dichotomous data were compared using the chi-square test. A P value < .05 was considered statistically significant. In instances of multiple comparisons, a Bonferroni correction was applied where the critical P value, $P_c = .05/(n\text{-}1)$, wherein "n" represents the total number of comparator groups. The geographic distribution of surgeon respondents was generated using Tableau Public v10.5 (Tableau Software, Inc. 2017).

Bellwether designation

The classification of bellwether status was based upon two factors: (1) the level of hospital designation advised by survey respondents, and (2) whether the procedure was deemed essential by both neurosurgeons and non-neurosurgeons. A majority (> 50%) of respondents had to agree that the procedure was best suited for a level 1 or level 2 healthcare facility. Procedures for which the majority believed should be performed at a level 3 facility only did not reach bellwether status. Hospital level definitions for low- and middle-income countries have been outlined by the World Bank and endorsed by the World Health Organization's (WHO) Emergency and Essential Surgical Care (EESC) guide [8]. A level 1 hospital, often termed a district, rural, or community hospital, is one offering mainly internal medicine, obstetrics and gynecology, pediatrics, and general surgery, or simply general practice. Level 2 hospitals are usually more differentiated by function with less than 10 specialties and are known also as regional or provincial hospitals. A level 3 hospital contains highly specialized staff and technical equipment including intensive care resources and advanced imaging capabilities. National, central, and academic teaching hospitals often qualify as level 3 facilities [8].

Respondents indicated the degree to which a procedure was deemed essential by rating the procedure on a continuous scale from 0 to 100, where 0 represents "insignificant; not necessary for surgeon to confidently perform," and 100 represents "essential; surgeon must be skilled in this procedure." A mean score of 90 or above was considered to indicate an essential procedure. To reach bellwether status, a procedure must have been deemed essential by both the cohort of neurosurgeons and the cohort of general surgeons involved in neurosurgical care.

Results

Study sample characteristics

Answers were obtained from 459 surgeons from 76 countries, including 369 neurosurgeons and 90 general pediatric surgeons (Table 1, Fig. 1). Among non-neurosurgeons, neurosurgical conditions comprised an average of 14% of their practice and volume. Forty-three percent of general surgeons reported routinely providing emergency neurosurgical care for children. Meanwhile, non-neurosurgical conditions accounted for an average of only 8% of the clinical volume of neurosurgeons worldwide. Across all regions, neurosurgeons reported performing an average of 216 cases per year (median, 200), while the mean case volume for general pediatric surgeons was significantly greater at 421 cases per year (median, 300) ($P < .001$).

The great majority of neurosurgeon and general surgeon respondents (88%) reported practicing primarily in a tertiary, comprehensive referral center, a feature that did not differ across WHO regions or income groups. Ninety-one percent of respondents' time distribution was either purely clinical (19%) or mostly clinical with some administrative duties (72%). Sixty-four percent (64%) of surgeons stated they worked in public hospitals, while 12% worked exclusively in private practice and 23% had a mixed practice in both public and private sectors.

Commonly performed procedures

The most commonly selected procedure was VPS, selected by 94% of respondents (Fig. 2). Next, 93% of respondents indicate commonly operating on posterior fossa tumors, a figure that did not significantly differ across World Bank income groups. A significant difference was observed between LIC and HIC in the frequency of performing endoscopy (60 vs. 91%, $P < .001$), craniofacial reconstruction (20 vs. 73%, $P < .001$), epilepsy surgery (0 vs. 62%, $P < .001$), vascular neurosurgery (20 vs. 72%, $P = .002$), and spinal instrumentation (40 vs. 71%, $P = .008$). Across World Bank income groups, there was no difference in how commonly was performed VPS (> 93%), trauma neurosurgery (> 74%), or brain tumor surgery (> 91%) (Fig. 2).

Table 1 Respondent characteristics

	Number (%)
Surgeon respondents	459 (100)
Neurosurgeons	369 (80.4)
Pediatric neurosurgeons	319 (69.5)
General pediatric surgeons	90 (19.6)
Neurosurgeon demographics ($n = 369$)	
LIC	5 (1.4)
LMIC	88 (23.9)
UMIC	63 (17.1)
HIC	213 (57.7)
AFR	13 (3.5)
AMR-L	40 (10.8)
AMR-US/C	57 (15.5)
EMR	25 (6.8)
EUR	90 (24.4)
SEAR	75 (20.3)
WPR	69 (18.7)

Bellwether procedures

Six pediatric neurosurgical procedures were identified as bellwether procedures according to the outlined criteria: ventriculoperitoneal shunt for hydrocephalus (VPS), myelomeningocele closure, burr holes, trauma craniotomy, external ventricular drain (EVD) insertion, and cerebral abscess evacuation (Table 2). Each of these procedures were independently considered essential by the collective group of neurosurgeon and general surgeon respondents (Fig. 3) and were considered necessary procedures for level 1 or level 2 healthcare centers (Fig. 4). The remaining nine candidate procedures did not meet bellwether criteria; however, all were deemed suitable for level 3 facilities (Fig. 5). Three procedures—posterior fossa tumor resection, complex spinal dysraphism repair, and Chiari decompression—were designated "essential" by neurosurgeon respondents, but not by general surgeons. Conversely, general surgeon respondents labeled spinal column fixation an essential pediatric neurosurgery procedure, but the mean score from the neurosurgeon cohort fell short of "essential" designation. While commonly performed at respondent hospitals, neither epilepsy nor craniofacial reconstruction was considered essential; surgical treatment for spasticity also was deemed non-essential by survey respondents.

Differences between bellwether procedure designation across World Bank income groups and WHO regions were explored. For VPS, burr holes, trauma craniotomy, and abscess evacuation, there was uniform agreement by respondents from all regions and groups regarding bellwether status (Table 3). While there was consensus among respondents on the essential nature of performing a posterior fossa tumor resection by neurosurgeons, its designation as a "level 3 facility only" procedure disqualified it from bellwether status. A greater proportion of respondents in Africa (98%) and low-income countries (LICs) (100%) considered myelomeningocele repair essential relative to those from high-income countries (HICs) (85%) and other WHO regions, including the Western Pacific region (82%) ($P < .005$) (Table 3). Similarly, endoscopic third ventriculostomy (ETV) was considered essential by more

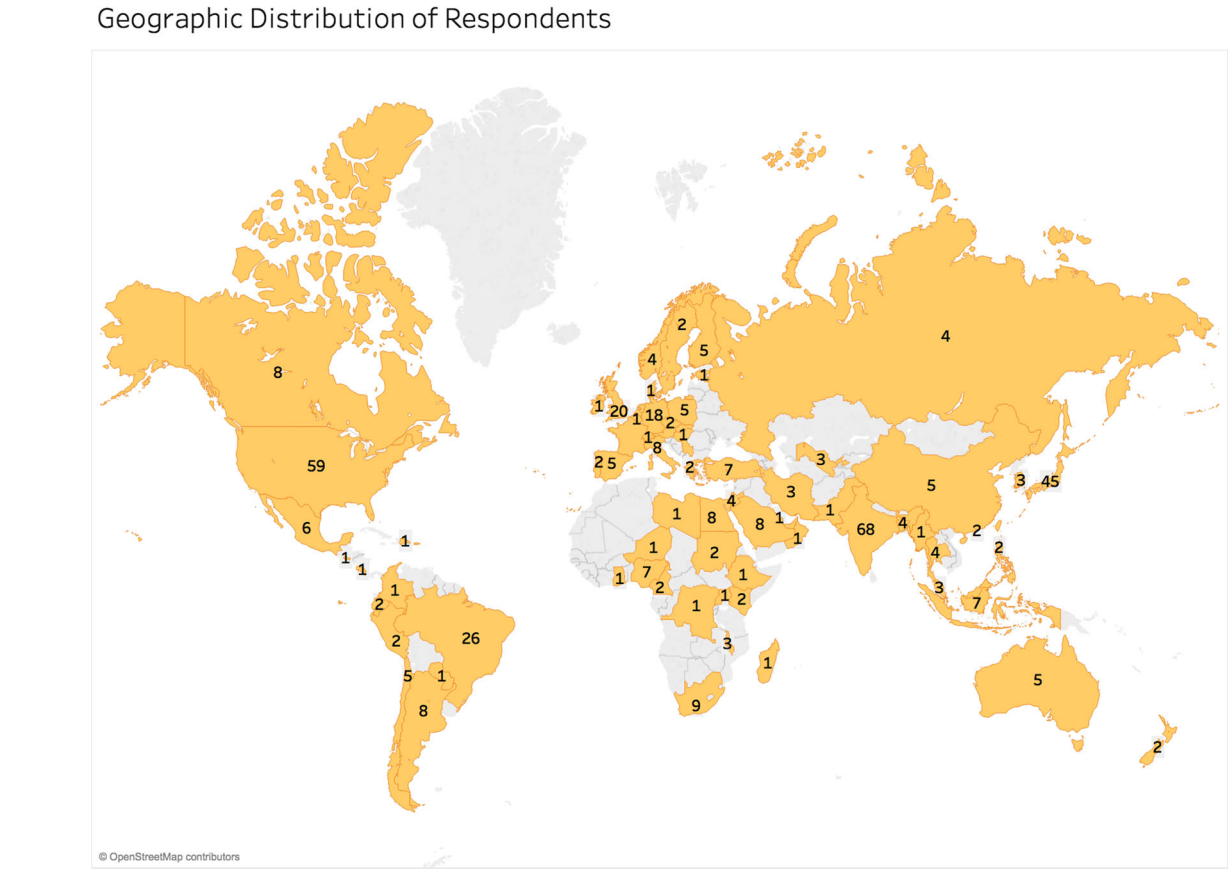

Fig. 1 Global map representing geographic location in which respondents primarily offer surgical care. Numbers indicate the number of respondents within each country

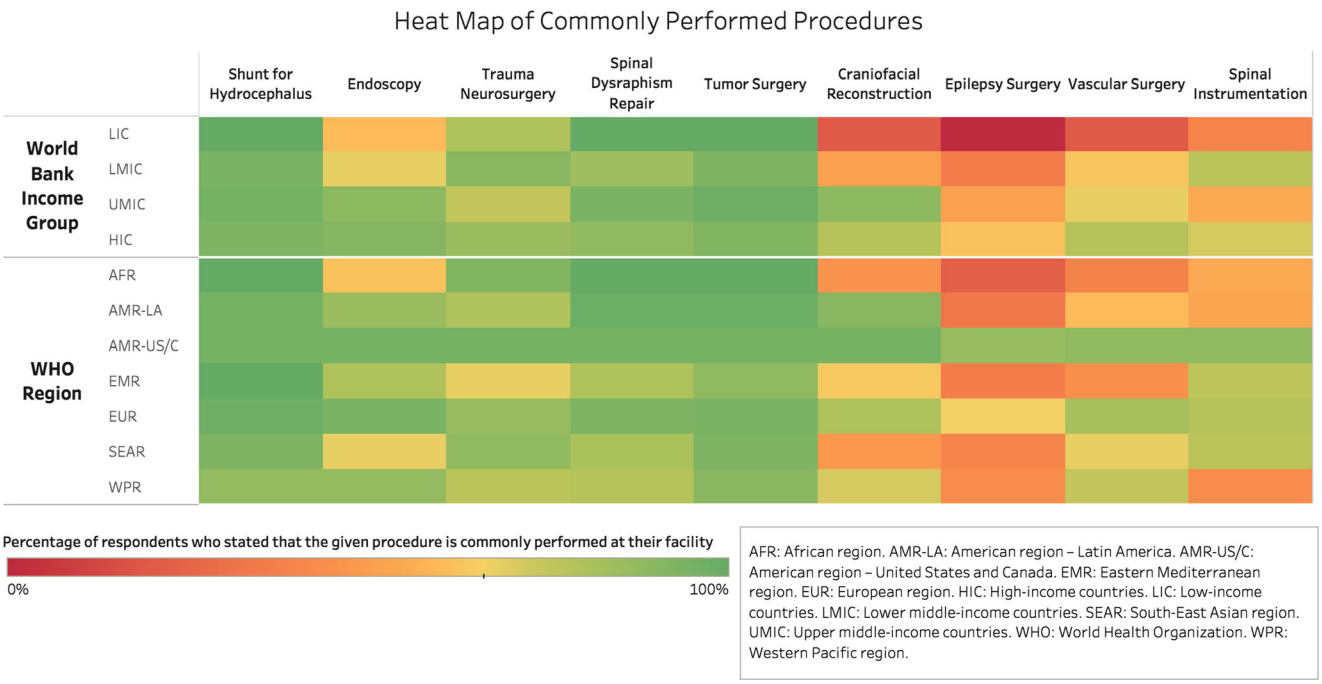

Fig. 2 Commonly performed procedures are indicated on a continuous color scale, organized by the World Bank income group and WHO region. The color gradient indicates percentage of respondents, not numeric case volume. Procedures commonly performed by a large percentage of respondents are indicated in dark green; those commonly performed by a lesser percentage are represented in dark red

Table 2 Bellwether designations

Procedure	% selecting Level 1 or Level 2 hospital designation	Deemed essential by neurosurgeons*	Deemed essential by general surgeons*	% stating procedure is commonly performed at their center	Classification
Ventriculoperitoneal shunt insertion	77	Yes	Yes	94	Bellwether
Myelomeningocele closure	52	Yes	Yes	88	
Burr holes for hematoma evacuation	85	Yes	Yes	84	
Trauma craniotomy	81	Yes	Yes	84	
External ventricular drain insertion	85	Yes	Yes	NA	
Cerebral abscess evacuation	69	Yes	Yes	NA	
Endoscopic third ventriculostomy	36	Yes	Yes	85	Non-Bellwether
Posterior fossa tumor resection	21	Yes	No	93	
Spinal cord detethering	30	Yes	Yes	88	
Complex dysraphism repair	16	Yes	No	NA	
Spinal column fixation	45	No	Yes	69	
Epilepsy surgery	7	No	No	52	
Craniofacial reconstruction	13	No	No	73	
Chiari malformation surgery	39	Yes	No	NA	
Surgery for spasticity	20	No	No	NA	

*"Essential" = score ≥ 90 on the essential rating scale from 0 to 100

respondents from LICs than from HICs (93 vs. 79%, $P = .006$).

Discussion

Herein, we propose a classification of bellwether procedures for the field of pediatric neurosurgery by means of an international survey of neurosurgeons and other pediatric surgical providers. A bellwether should address substantial need, be cost-effective, and be feasible to implement [9]. Six neurosurgical procedures were deemed essential within the neurosurgeon's skillset *and* necessary for level 1 or level 2 healthcare facilities: VPS, myelomeningocele closure, burr holes, trauma craniotomy, EVD insertion, and cerebral abscess evacuation. At a minimum—and in order to provide more equitable delivery of surgical care—level 2 healthcare facilities and above, in every country worldwide, should have the ability to safely and effectively offer these six procedures to patients. By focusing on high-impact procedures, healthcare systems with limited resources will be able to maximize value-based care, thereby saving lives and minimizing disease-related disability.

Bellwether procedures are fundamental surgical procedures that predict accomplishment of many other essential surgical procedures and, thereby, serve as a metric by which to gauge adequacy of surgical delivery in a given community, country, or region [6]. In their original piece designed to gain a broad understanding of all surgical care, O'Neill et al. considered three bellwether procedures: laparotomy, caesarian delivery, and treatment of open fractures. Using the WHO EESC Global database, the authors found that performing these three procedures correlated well with the ability to perform many other important general, obstetric, and orthopedic procedures. Of note, neurosurgical procedures were not examined in their analysis. Designating bellwethers establishes treatment priorities, guides resource allocation, and facilitates data collection and objective evaluation of performance [10]. Inclusion of pediatric neurosurgical bellwethers within a Ministry of Health's National Surgical Plan will be an early and essential step in achieving universal pediatric neurosurgical coverage, particularly in resource-limited countries [11].

There exists no universally accepted methodology for designating bellwether procedures. Others have compiled an expert panel to propose bellwether procedures and then queried international health databases to identify surgical activity [6]. For neurosurgery—and pediatric neurosurgery in particular—there exists no reliable international database to capture surgical type or volume. Instead, we solicited the expertise of hundreds of surgical providers across all continents and income levels. To avoid a myopic interpretation of surgical priorities, non-neurosurgeons were included among the survey respondents. For a given procedure to attain bellwether status, general surgeons had to agree on its essential nature and the level of hospital within which it should be performed.

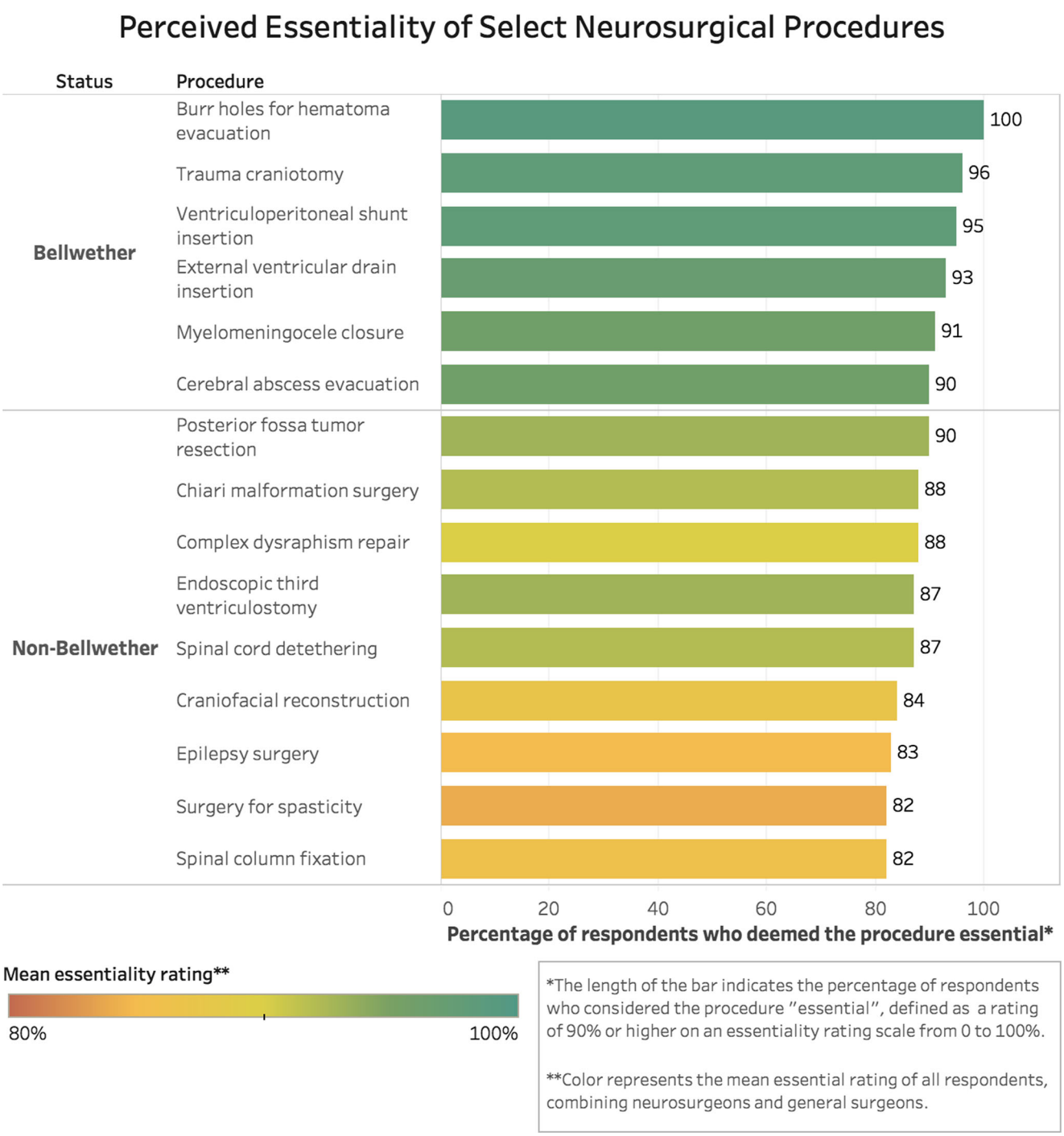

Fig. 3 Designation of essential neurosurgical procedures by neurosurgery and general surgery providers. Each procedure is shaded according to the average essentiality rating ascribed by respondents. The horizontal axis represents the percentage of respondents who considered the procedure "essential" by numerically grading the procedure at or above 90 on a 100-point scale

As expected, most of the proposed neurosurgical procedures were not designated as essential procedures for a level 1 or level 2 hospital, including posterior fossa tumor resection, ETV, epilepsy surgery, and spinal column fixation. While no less important, these more complex procedures can be centralized via referral to larger, level 3 centers with greater resources capable of offering complex surgical care safely [12, 13]. This paradigm is appropriate for high-income and low-income countries alike and has been shown to yield superior patient outcomes across numerous neurosurgical diseases [14–16].

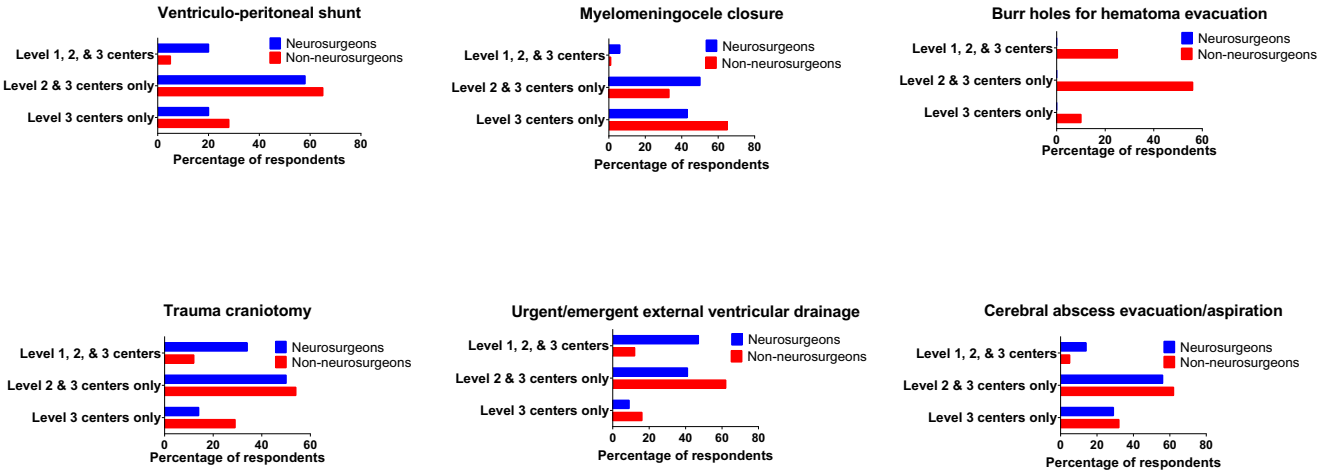

Fig. 4 Hospital designation by respondents for bellwether procedures. To achieve bellwether status, 50% or more of respondents had to select level 1 or level 2 designation

Several interesting observations from this survey warrant elaboration. ETV, which fell short of bellwether designation, was deemed essential by 93% of respondents in LIC and lower-middle income countries (LMIC), relative to 79% in high-income countries. The greatest incidence of childhood hydrocephalus emanates from LIC/MICs—especially in Africa, Latin America, and South East Asia [17]—where ETV has been shown to be an effective alternative to permanent implant-regulated CSF diversion [18, 19]. To minimize morbidity associated with shunt infection and malfunction, it may be considered, for both financial and clinical reasons, more important for neurosurgeons in LIC/MICs to safely perform ETV. Similarly, the surgical repair of myelomeningocele, which occurs at a much higher incidence in LIC/MICs [1], was felt to be essential by 100% of respondents from LICs relative to 85% in HICs ($P = .001$). In general, respondents from lower-income countries were more inclined to label procedures as essential, even if the procedures did not meet bellwether criteria by the collective group. Such procedures included epilepsy surgery, complex dysraphism repair, spinal column fixation, craniofacial reconstruction, and surgery for spasticity. One potential explanation for this is that surgeon respondents in LIC/MICs may represent the only neurosurgical providers for an entire region or country; if they are not capable of safely offering these services, an entire population will be left without proper care.

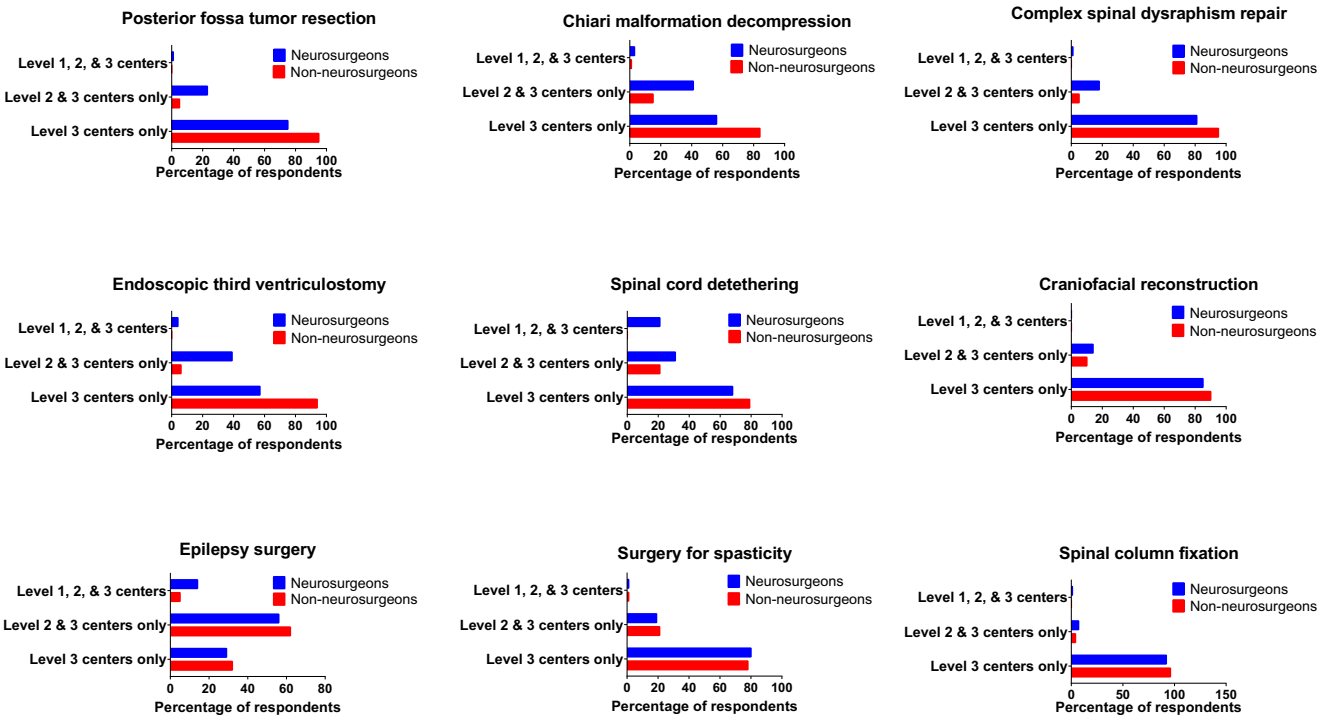

Fig. 5 Hospital designation by respondents for procedures not attaining bellwether status

Table 3 Essential rating: heterogeneity across income groups and WHO regions

Procedure	Percentage deemed essential by all respondents*	Mean essential rating** (0 = insignificant—> 100 = essential)	Heterogeneity across: World Bank income groups ($P = .017$)***	Heterogeneity across: WHO regions ($P = .0083$)****	Details
Ventriculoperitoneal shunt insertion	95	98	None ($P = 0.018$)	None ($P = 0.479$)	
Myelomeningocele closure	91	96	Yes ($P = 0.001$)	Yes ($P = .004$)	100% in LIC consider MMC essential relative to 85% in HIC ($P = .001$); 98% in AFR consider MMC essential relative to 82% in WPR ($P = .004$)
Burr holes for hematoma evacuation	100	99	None ($P = 1.0$)	None ($P = 1.0$)	
Trauma craniotomy	96	98	None ($P = .157$)	None ($P = .114$)	
External ventricular drain insertion	93	97	None ($P = .28$)	Yes ($P = .017$)	98% in WPR consider acute EVD insertion essential relative to 80% in SEAR ($P = .017$)
Cerebral abscess evacuation	90	96	None ($P = .228$)	None ($P = .162$)	
Endoscopic third ventriculostomy	87	93	Yes ($P = .006$)	None ($P = .640$)	93% in LIC consider ETV essential relative to 79% in HIC ($P = .006$)
Posterior fossa tumor resection	90	93	None ($P = .108$)	None ($P = .135$)	
Spinal cord detethering	87	92	None ($P = .112$)	None ($P = .162$)	93% in LIC consider complex dysraphism repair essential relative to 81% in HIC ($P = .015$)
Complex dysraphism repair	88	90	Yes ($P = .015$)	None ($P = .098$)	97% in LIC consider spinal column fixation essential relative to 69% in HIC ($P < .001$); Relative to more than 94% of respondents in AFR and AMRLA, 73% of respondents in AMRUSC ($P < .001$) and 66% of respondents in WPR ($P < .001$) consider spinal fixation essential.
Spinal column fixation	82	86	Yes ($P < .001$)	Yes ($P < .001$)	
Epilepsy surgery	83	85	Yes ($P < .001$)	Yes ($P = .009$)	92% in LMIC consider epilepsy surgery essential relative to 74% in HIC ($P < .001$); 95% in EMR consider epilepsy surgery essential relative to 68% in WPR ($P = .009$)
Craniofacial reconstruction	84	87	Yes ($P = .001$)	None ($P = .043$)	93% in LIC consider craniofacial repair surgery essential relative to 75% in HIC ($P = .001$)
Chiari malformation surgery	88	92	Yes ($P = .002$)	None ($P = .093$)	94% in LMIC consider Chiari decompression surgery essential relative to 80% in HIC ($P = .002$)
Surgery for spasticity	82	84	Yes ($P < .001$)	Yes ($P = .002$)	93% in LIC consider spasticity surgery essential relative to 69% in HIC ($P < .001$); 91% in EMR consider spasticity surgery essential relative to 66% in WPR ($P = .002$)

*"Essential" = score >/= 90 on the essential rating scale from 0 to > 100

**Combining both neurosurgeon and general surgeon responses

***Bonferroni's correction: .05/3 = .01666

****Bonferroni's correction: .05/6 = .0083

The implications of this study entail a significant responsibility to healthcare systems worldwide. The requisite nature of these six neurosurgical procedures by level 2 hospitals warrants a large-scale effort to significantly expand and enhance the neurosurgical workforce. Recent evidence suggests a significant deficit in neurosurgical providers in LIC/MICs irrespective of hospital level designation [1]. But while the fiscal costs of capacity-building efforts will be substantial, the economic costs of neglected neurological disease are untenable, eclipsing four trillion USD in LIC/MICs by 2030 [20]. And while many child health programs ignore routine surgical conditions [3], effective and timely pediatric surgical care has been shown to be cost-effective in developing countries [21]. An additional annual global expenditure of approximately $3 billion USD used to equip primary-level facilities in LIC/MICs with essential surgery capabilities is estimated to generate a benefit-cost ratio of 10:1 [9].

The results outlined above must be considered within the context of several relevant limitations. The validity of any survey depends upon the experience and integrity of individual respondents. While bias was mitigated by including non-neurosurgeons, the neurosurgical-centric nature of the survey questions and answers render the data susceptible to responder bias. Acceptance and adoption of the proposed bellwether classification by the international healthcare community will likely require advocacy, time, and, ultimately, proof of clinical efficacy. Finally, because most surgical societies were unable to provide an accurate membership census, calculating the total number of survey invitations—and therefore the response rate—was not possible.

These limitations notwithstanding, the cumulative voice of more than 450 surgeons worldwide, produce a reliable depiction of which neurosurgical interventions are likely to most effectively avert childhood disability and death.

Conclusions

Six bellwether procedures for pediatric neurosurgery are identified via an international survey of surgical providers: VPS, myelomeningocele closure, burr holes, trauma craniotomy, EVD insertion, and cerebral abscess evacuation. ETV, posterior fossa tumor resection, and spinal column fixation were also deemed essential procedures by respondents but may be better suited for tertiary-level centers only. Hospitals capable of performing bellwether procedures are likely to offer other essential neurosurgical procedures. Equipping primary- or secondary-level facilities with the resources to safely and effectively perform bellwether procedures will dramatically enhance the timely delivery of necessary neurosurgical care to children. The substantial investment in surgical capacity-building would be paralleled by significant economic gains by averting preventable disability and death.

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Compliance with ethical standards

Conflicts of interest None.

Abbreviations AFR, African region; AMR-LA, American region—Latin America; AMR-US/C, American region—United States and Canada; CT, computed tomography; EESC, Emergency and Essential Surgical Care; EMR, Eastern Mediterranean region; ETV, endoscopic third ventriculostomy; EUR, European region; HICs, high-income countries; LICs, low-income countries; LIC/MICs, low- and middle-income countries; LMICs, lower middle-income countries; MRI, magnetic resonance imaging; SEAR, South-East Asian region; UMICs, upper middle-income countries; VPS, ventriculoperitoneal shunt; WHO, World Health Organization; WPR, Western Pacific region

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