I

t is by now well recognized that safe and affordable surgical care is not available to the majority of the world’s population.1 A significant subset of this deficit is accounted for by the absence of neurosurgical expertise and, specifically, pediatric neurosurgical expertise in low- and middle-income countries (LMICs).3 The response of the pediatric neurosurgical community has been noteworthy, with as many as half of surveyed pediatric neurosurgeons, indicating consistent annual involvement in international neurosurgical efforts.4 Additionally, the implementation of international courses and digital platforms has increased the frequency and availability of international neurosurgical engagement. While quantity is an important first step, it will become increasingly important to also evaluate the quality and clinical outcomes of such engagements. Currently, there are few published reports of surgical correction for craniosynostosis in Vietnam.7–10 As such, we describe the initial establishment of a pediatric craniosynostosis center in southern Vietnam with initial surgical outcomes.

Nonsyndromic craniosynostosis in Vietnam: initial surgical outcomes of subspecialty mentorship

Dang Do Thanh Can, MD,1,2 Jacob R. Lepard, MD,3,4 Nguyen Minh Anh, MD,1 Pham Anh Tuan, MD,1 Tran Diep Tuan, MD,1 Vo Tan Son, MD,1 John H. Grant, MD,3 and James M. Johnston, MD3,4

1University of Medicine and Pharmacy at Ho Chi Minh City, Vietnam; 2Neurosurgical Department, Children’s Hospital 2, Ho Chi Minh City, Vietnam; 3Department of Neurological Surgery, University of Alabama at Birmingham, Birmingham, Alabama; 4Section of Pediatric Neurosurgery, Children’s of Alabama, Birmingham, Alabama; and 5Department of Plastic Surgery, University of Alabama at Birmingham, Birmingham, Alabama

OBJECTIVE There is a global deficit of pediatric neurosurgical care, and the epidemiology and overall surgical care for craniosynostosis is not well characterized at the global level. This study serves to highlight the details and early surgical results of a neurosurgical educational partnership and subsequent local scale-up in craniosynostosis correction.

METHODS A prospective case series was performed with inclusion of all patients undergoing correction of craniosynostosis by extensive cranial vault remodeling at Children’s Hospital 2, Ho Chi Minh City, Vietnam, between January 1, 2015, and December 31, 2019.

RESULTS A total of 76 patients were included in the study. The group was predominantly male, with a male-to-female ratio of 3.3:1. Sagittal synostosis was the most common diagnosis (50%, 38/76), followed by unilateral coronal (11.8%, 9/76), bicoronal (11.8%, 9/76), and metopic (7.9%, 6/76). The most common corrective technique was anterior cranial vault remodeling (30/76, 39.4%) followed by frontoorbital advancement (34.2%, 26/76). The overall mean operative time was 205.8 ± 38.6 minutes, and the estimated blood loss was 176 ± 89.4 mL. Eleven procedures were complicated by intraoperative durotomy (14.5%, 11/76) without any damage of dural venous sinuses or brain tissue. Postoperatively, 4 procedures were complicated by wound infection (5.3%, 4/76), all of which required operative wound debridement. There were no neurological complications or postoperative deaths. One patient required repeat reconstruction due to delayed intracranial hypertension. There was no loss to follow-up. All patients were followed at outpatient clinic, and the mean follow-up period was 32.3 ± 18.8 months postoperatively.

CONCLUSIONS Surgical care for pediatric craniosynostosis can be taught and sustained in the setting of collegial educational partnerships with early capability for high surgical volume and safe outcomes. In the setting of the significant deficit in worldwide pediatric neurosurgical care, this study provides an example of the feasibility of such relationships in addressing this unmet need.

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KEYWORDS global neurosurgery; craniosynostosis; Vietnam; limited resource; pediatric neurosurgery; craniofacial
Methods

Patient Selection

Approval for this study was obtained from the ethics review boards at the University of Medicine and Pharmacy at Ho Chi Minh City, Children’s Hospital 2 (CH2), Vietnam, and the University of Alabama at Birmingham. All children younger than 18 years undergoing surgical correction of craniosynostosis were included in the case series. Patients were excluded if they had a known diagnosis or phenotypic features suggestive of a congenital syndrome contributing to craniofacial deformity. All diagnoses of syndromic versus nonsyndromic craniosynostosis were made based on history and physical examination findings due to the unavailability of confirmatory genetic testing. The study period included all patients undergoing surgery between January 1, 2015, and December 31, 2019. All data were collected prospectively.

Subspecialty Mentorship

The first author (D.D.T.C.) underwent neurosurgical residency training at the University of Medicine and Pharmacy at Ho Chi Minh City and Cho Ray Hospital, with supervision from the local senior authors prior to founding a new pediatric neurosurgical department at CH2 in 2011. All procedures were performed by the first author and occurred at CH2, which is a large pediatric tertiary care center in Ho Chi Minh City, southern Vietnam. The neurosurgical department at this hospital consists of 10 pediatric neurosurgeons and performs 600 neurosurgical procedures each year and 500 neurosurgical outpatient visits per week. Broad access to general pediatric care exists in Vietnam, with 99% of infants receiving vaccinations according to WHO recommendations. The initial diagnoses of craniosynostosis are made at the local health center and hospital level by general practitioners and pediatricians based on abnormal cranial shape with further referral to tertiary neurosurgical centers such as CH2.

All patients underwent cranial CT with 3D reconstruction prior to surgical correction to confirm the diagnosis and allow for collaborative surgical planning. A single dose of cephazolin was given prior to incision and at closing for each surgical case. Rigid interosseous fixation was achieved using absorbable cranial plates in nearly all cases in this series. Titanium plates or suture were only used in the rare circumstance in which absorbable implants were unavailable. Postoperatively, all patients were admitted overnight to the pediatric intensive care unit and were subsequently transferred to the neurosurgical ward. Wound care and evaluation were performed daily by ward nursing staff and during daily physician ward rounds. Between 10 and 20 new patients with craniosynostosis are treated at CH2 annually.

The initial partnership between CH2 and the Children’s of Alabama (COA) Global Surgery Program was initiated in 2013 and has been described in detail elsewhere. As a component of this partnership, craniofacial surgeries were performed during each annual visit, with the first author acting as the primary surgeon and the visiting surgeons providing instruction as needed. In addition to this, the first author participated in a 3-month visiting training fellowship at COA, during which time additional exposure was gained in various operative techniques and treatment strategies for craniosynostosis (Fig. 1). In the interval between visits, consistent telecollaboration was maintained in the form of email exchange with discussion of patients and assistance with formulation of operative strategies for craniofacial correction (Fig. 2). Prior to this partnership, surgical correction for pediatric craniosynostosis occurred rarely and without consistency in the largest Vietnamese neurosurgical centers, often depending on the presence of international volunteers or travel abroad for neurosurgical evaluation.

Statistical Analysis

Data were collected prospectively between January 1, 2015, and December 31, 2019. Patient demographic, clinical, perioperative, operative, and outcomes data were re-
corded in SPSS version 26 (IBM Corp.). Aesthetic outcomes were assigned by the primary surgeon at 6 months and each subsequent year at postoperative follow-up according to the classification system of Aryan et al. There was no independent rater for aesthetic ratings. Descriptive statistics were used to summarize clinically relevant trends and patient characteristics.

Results
Demographics
A total of 76 patients were included in the study. The group was predominantly male, with a male-to-female ratio of 3.3:1. The median age at presentation for neurosurgical evaluation was 17 months (IQR 11–30 months) with a range of 4 to 96 months. Sagittal synostosis was the most common diagnosis (50%, 38/76), followed by unilateral coronal (11.8%, 9/76), bicoronal (11.8%, 9/76), and metopic (7.9%, 6/76). There were 4 patients with lambdoid suture involvement, 2 with unilateral (2.6%, 2/76) and 2 with bilateral (2.6%, 2/76). Another 10 patients (27.1%, 10/76) had combined involvement of multiple sutures. The male predominance was notable in the patients with sagittal synostosis (male-to-female ratio 6.1:1), com-

FIG. 2. Examples of telecollaboration for presurgical planning with osteotomy recommendations provided by the plastic surgery mentor in the United States for cases of sagittal and bilateral lambdoid synostoses (A) and operative photographs for the same case performed independently in Vietnam (B). Similarly, osteotomy recommendations for case of unicoronal + sagittal synostosis (C) and operative photographs prior to closure (D) are shown. Figure is available in color online only.
bined suture involvement (male-to-female ratio 9:1), and metopic synostosis (male-to-female ratio 5:1). There was no sex predilection for coronal or lambdoid synostosis (Table 1).

Length of Stay and Surgical Technique

All patients were admitted to the inpatient ward prior to surgery, with surgical delay dependent on operating room availability. The median inpatient stay prior to surgery was 7 days (IQR 3–17 days) with an overall range of 1 to 40 days. Postoperatively, patients had an inpatient stay for a median of 9 days (IQR 8–14.8 days) with an overall range of 4 to 36 days. Thirty patients underwent anterior two-thirds cranial vault remodeling (CVR) (Fig. 3), and 26 patients underwent frontoorbital advancement (FOA). Another 3 patients underwent FOA with biparietal expansion, all 3 of whom had unicoronal and sagittal combined synostoses. Seven patients underwent posterior cranial vault remodeling. Three patients underwent a Pi technique craniectomy, and another 3 patients underwent total cranial remodeling (Table 2).

Operative Time, Complications, and Outcomes

The overall mean operative time for all surgical tech-
niques was 205.8 ± 38.6 minutes. The mean perioperative blood transfusion for all surgical techniques was 176 ± 89.4 mL, with 90.8% (69/76) of patients requiring transfusion within the intraoperative or immediate postoperative period (Table 3). In total, 11 procedures were complicated by duretomy (14.5%, 11/76), which happened more frequently with FOA (7/11) than with anterior CVR (2/11) and posterior CVR (2/11). There were no further intraoperative complications such as injury to dural venous sinuses, subdural hemorrhage, or cerebral contusion.

Postoperatively, 4 procedures were complicated by wound infection (5.3%, 4/76), all of which required operative wound debridement. Three wound infections occurred after FOA for coronal synostosis, and 1 occurred after CVR for sagittal synostosis. One patient developed postoperative meningitis (1.3%, 1/76) following FOA, which was successfully treated with 2 weeks of intravenous antibiotics without further sequelae. Another patient developed an intracranial abscess after posterior CVR, requiring operative evacuation and removal of all implants due to infectious complications. Two other patients required additional remodeling. One developed a wound infection resulting in a poor aesthetic outcome requiring revision. Another patient initially underwent a Pi procedure at age 7 months and subsequently developed delayed intracranial hypertension after 3 years diagnosed by papilledema and diffuse copper-beaten cranium (Fig. 4). There were no permanent neurological complications or postoperative deaths in our series.

In all, there were 5 patients who required repeat operation due to infectious complications. Two other patients required additional remodeling. One developed a wound infection resulting in a poor aesthetic outcome requiring revision. Another patient initially underwent a Pi procedure at age 7 months and subsequently developed delayed intracranial hypertension after 3 years diagnosed by papilledema and diffuse copper-beaten cranium (Fig. 4). There were no permanent neurological complications or postoperative deaths in our series.

No patients were lost to follow-up, and the average follow-up period was 32.3 ± 18.8 months postoperatively. The classification of aesthetic outcomes established by Aryan et al. was used in this study.27 A total of 40 patients (52.6%, 40/76) were rated as having grade 1 aesthetic outcomes at the final follow-up indicating excellent correction with no visible irregularity. Twenty-four patients (31.6%, 24/76) were rated as having grade 2 outcomes indicating good correction with minimal irregularity. Nine patients were rated as having grade 3 outcomes (11.8%, 9/76) indicating a compromised correction but not requiring repeat operation. Lastly, 3 were rated as having grade 4 aesthetic outcomes, indicating compromised correction requiring repeat operation (3.9%, 3/76). Patients with sagittal and metopic synostosis had better aesthetic outcomes with more than 85% excellent and good results, whereas approximately one-third of patients with unicoronal synostosis or bicoronal synostosis had grade 3 or 4 aesthetic results.

Discussion

This study describes the initial surgical outcomes of a pediatric craniosynostosis center in southern Vietnam. These data provide important insight into the early outcomes of international neurosurgical mentorship and surgical management of craniosynostosis in a middle-income country (MIC) and demonstrate epidemiological trends for craniosynostosis in southeast Asia.

International Mentorship

The experience reported here demonstrates a relatively rapid scale-up of surgical correction for craniosynostosis in a setting where none was previously being undertaken. This expedited growth was largely possible due to the very high level of technical expertise already present among the Vietnamese surgical team, such that all that was needed was focused teaching on operative decision-making and details of the surgical technique. Additionally, given that CH2 is a major tertiary referral center, there was already an economy of scale present in the form of high-level anesthesia support and neurosurgical ancillary staff; thus, the perioperative infrastructure for performing these cases was already in place.

In our series, the average operative time for surgical correction was 205.8 minutes, and the periprocedural blood transfusion rate was 90.8% (69/76) with a mean transfusion volume of 176 mL, which is comparable to many series published from higher-income settings.18–20 The rate of reoperations for infectious complications was higher in our study than in similar North American21,22 and Chinese23 studies. There is a paucity of literature regarding complication rates of craniosynostosis correction in LMICs; however, it is possible that this finding is a component of the learning curve in adopting a new surgical technique as well as an environmental risk factor of performing complex cranial procedures in the LMIC setting in which higher rates of surgical site infections are well established.24 Overall, the potential for this rapid scale-up with relatively good surgical outcomes highlights an important nuance in setting appropriate expectations for global neurosurgical partnerships—while low-income countries (LICs) and MICs tend to be lumped together into the single cat-

### TABLE 2. Mean, median, and ranges of age at presentation and number of hospitalization days pre- and postsurgery

<table>
<thead>
<tr>
<th>Age at Presentation (mos)</th>
<th>Preop Stay (days)</th>
<th>Postop Stay (days)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean ± SD</td>
<td>23.5 ± 18.9</td>
<td>11.1 ± 10.8</td>
</tr>
<tr>
<td>Median (IQR)</td>
<td>17 (11–29.75)</td>
<td>7 (3–17)</td>
</tr>
<tr>
<td>Range</td>
<td>4–96</td>
<td>1–40</td>
</tr>
</tbody>
</table>

### TABLE 3. Operative time and perioperative transfusion volume by type of corrective procedure

<table>
<thead>
<tr>
<th>Procedure</th>
<th>Op Time (mins)</th>
<th>Transfusion Vol (mL)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Anterior cranial remodeling (n = 30)</td>
<td>185.7 ± 21.7</td>
<td>166.7 ± 75.8</td>
</tr>
<tr>
<td>FOA (n = 26)</td>
<td>223.7 ± 28.8</td>
<td>206.7 ± 86.2</td>
</tr>
<tr>
<td>FOA + biparietal expansion (n = 3)</td>
<td>291.7 ± 16.1</td>
<td>208.3 ± 72.2</td>
</tr>
<tr>
<td>Pi technique (n = 3)</td>
<td>166.7 ± 11.5</td>
<td>41.7 ± 72.2</td>
</tr>
<tr>
<td>Posterior cranial expansion (n = 7)</td>
<td>164.3 ± 13.7</td>
<td>107.1 ± 112.5</td>
</tr>
<tr>
<td>Total cranial remodeling (n = 7)</td>
<td>247.1 ± 26.1</td>
<td>214.3 ± 61</td>
</tr>
<tr>
<td>Total (n = 76)</td>
<td>205.8 ± 38.6</td>
<td>176 ± 89.4</td>
</tr>
</tbody>
</table>

Values represent mean ± SD.
egory of “LMICs,” there are often vast differences in the level of baseline neurosurgical capability between the two. According to the most recent estimates, Vietnam has approximately 600 neurosurgeons with a neurosurgeon density of 6.6 per 1 million population. By comparison, neighboring Laos has only 3 neurosurgeons and Cambodia has 25 neurosurgeons with densities of 0.45 and 1.6 per million population, respectively. Similarly, as of 2016, there were an estimated 488 neurosurgeons in all of sub-Saharan Africa, with an average density of 0.51 neurosurgeons per 1 million population. In contexts such as these, all of which are categorized by the World Bank as LICS, the neurosurgeons who are available are more likely to be overwhelmed by the higher-volume and more urgent pathologies such as trauma, hydrocephalus, and spinal dysraphism. The relatively high number of neurosurgeons in Vietnam, which is an MIC, has allowed for the luxury of subspecialization and provided an ideal opportunity for focused subspecialty mentorship in specific areas of interest such as craniosynostosis. This experience should emphasize to the pediatric neurosurgeon who is interested in global neurosurgical education that the local context is highly variable by country and should be a consideration in setting appropriate goals and expectations for partnerships.

It is worth considering that several external factors also played a significant role in the rapid development of this Vietnamese craniofacial center. For instance, Vietnam is in the process of implementing a universal health coverage system, which prevents patients from shouldering much of the financial burden of care. Such systems are not common in other low-income regions and represent an additional barrier to accessing care. Additionally, significant institutional support on the part of both CH2 and COA Global Surgery allowed funding and time allowance for extended and consistent travel without which the partnership would not have been a possibility.

**Epidemiological Trends**

The incidence of pediatric craniosynostosis is estimated to be approximately 1–2 per 10,000 live births, often with a male predominance; however, globally, the epidemiology is not well characterized. Kolar recently reported epidemiological characteristics of craniosynostosis based on a series of 690 patients in the United States. In their series, sagittal synostosis was the most common single-suture synostosis followed by metopic synostosis, represented by approximately one-half and one-quarter of patients, respectively. Both of these midline synostoses had a strong male predominance while the coronal synostoses had a female predominance. Similar trends were reported by Di Rocco et al. in a series of 2808 children with craniosynostosis in western Europe. In contrast to these western series, Kadri and Mawla reported on a series of 116 cases of craniosynostosis from Syria in which metopic synostosis was the most common diagnosis at 24% of the cohort followed by sagittal synostosis with 22% of patients. Interestingly, there has been some speculation as to whether ethnic variations in the epidemiology of craniosynostosis might exist. Anderson et al. compared a cohort of primarily Caucasian and Asian patients and found a significantly lower proportion of sagittal involvement in the Asian population. However, our series does not support this claim, given that 50% of our patients presented with isolated sagittal synostosis. Similarly, in a South Korean series, Byun et al. reported sagittal involvement as the most common diagnosis. While our single-center experience is insufficient to provide population-wide data and birth incidence per population, it does provide valuable insight into the proportional frequencies of the various synostoses.

**Increasing Access to Care**

A recent study by Mullapudi et al. estimated that only
3% of the pediatric population in LICs and 8% in MICs had access to safe and affordable surgical care. Large efforts have been made over the last several decades to provide pediatric surgical care via mission model organizations, particularly for craniofacial disease. However, such efforts are no replacement for a local neurosurgeon trained to treat pediatric neurosurgical disease who is then capable of training other local surgeons to do the same. This is particularly true of craniosynostosis in which one of the primary indications for surgery is improvement of aesthetic outcomes. Recent studies have demonstrated that significant variation exists in what is considered “normal” craniofacial morphology. It is likely that such subjective judgments would be far more difficult to negotiate in the setting of cross-cultural barriers, as is the case with visiting surgeons. Thus, a local “same-culture” neurosurgeon is likely to provide significant benefit in the treatment of craniosynostosis, in addition to the more intuitive benefits of local pediatric neurosurgical care such as decreased travel and financial expenditure, among others.

Notably, no prior literature exists regarding the surgical treatment of craniosynostosis in Vietnam. Prior to this endeavor in subspecialty training, these cases occurred only rarely in Vietnam and were dependent on the physical presence of international volunteers, otherwise requiring families to travel abroad to seek care. These prospectively collected data provide important insights into how craniosynostosis care can be taught and provided in MICs; however, it is limited to the experience of a single local surgeon and institution. In the context of the significant global deficit in pediatric neurosurgical expertise, we propose this example of mentorship and rapid increase in local surgical care as a feasibility study of which many similar examples are needed. Assuming a conservative birth incidence of 2 per 10,000 live births for craniosynostosis, based on 2018 population and birth rates, the country of Vietnam should have approximately 320 cases of craniosynostosis annually. Such volumes could conceivably be managed by a few national-level pediatric tertiary care centers around the country in the near future based on current local growth rates of neurosurgical expertise. However, making the same calculations for craniosynostosis in Vietnam can undergo local surgical treatment with satisfactory cosmetic results and without any severe morbidities or mortality. Surgical care for pediatric craniosynostosis can be taught and sustained in the setting of collegial educational partnerships with early capability for high surgical volume and safe outcomes. In the setting of the significant deficit in worldwide pediatric neurosurgical care, this model serves as an example of the feasibility of such relationships in addressing this unmet need.

Conclusions

We found that most patients with nonsyndromic craniosynostosis in Vietnam can undergo local surgical treatment with satisfactory cosmetic results and without any severe morbidities or mortality. Surgical care for pediatric craniosynostosis can be taught and sustained in the setting of collegial educational partnerships with early capability for high surgical volume and safe outcomes. In the setting of the significant deficit in worldwide pediatric neurosurgical care, this model serves as an example of the feasibility of such relationships in addressing this unmet need.

References


Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: all authors. Acquisition of data: Lepard. Drafting the article: Lepard. Critically revising the article: Johnston. Reviewed submitted version of manuscript: Can, Anh, PA Tuan, TD Tuan, Son, Grant, Johnston. Statistical analysis: Johnston. Reviewed submitted version of manuscript: Can, Anh. Study supervision: Can.

Correspondence
Jacob R. Lepard: University of Alabama at Birmingham, AL. jlepard@uabmc.edu.