Patterns of Hydrocephalus in Rural Haiti: A Computed Tomography–Based Study

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■ INTRODUCTION/OBJECTIVE: **Hydrocephalus is a common neurosurgical disorder that can lead to significant disability or death if not promptly identified and treated. Data on the burden of hydrocephalus in low-income countries are limited, given a lack of radiologic resources for the diagnosis of this condition. Here, we present an analysis of patterns of hydrocephalus from a large sample of computed tomography (CT) scans of the head performed at a public hospital in rural Haiti, a low-income country in the Caribbean.**

■ METHODS: **We analyzed reports from 3614 CT scans of the head performed between July 2013 and January 2016 for findings that were consistent with a diagnosis of hydrocephalus (report indicating "hydrocephalus," "ventriculomegaly," or "enlargement of the ventricles"). Extracted data included demographics, study indication, radiologic findings, and reported etiology of hydrocephalus.**

■ RESULTS: **In total, 119 scans had findings concerning for hydrocephalus (3.5% of all scans, 6.3% of abnormal scans; age range 0–90 years; median age 35.5 years; 49.6% male). Pediatric patients (<18 years of age) accounted for 39% of cases. In total, 113 of 119 (95%) scans had indications for possible neurosurgical intervention. Among these 113 scans, 36 (30%) scans demonstrated communicating hydrocephalus, 66 (55%) scans demonstrated noncommunicating hydrocephalus (primarily due to intraventricular hemorrhage [27 scans, 23%] or brain tumors [24, 20%]), and 11 (9%) scans were indeterminate regarding whether the hydrocephalus was communicating versus noncommunicating.**

■ CONCLUSIONS: **In a large sample of CTs performed in a rural low-income setting, hydrocephalus was common, predominantly noncommunicating, and often associated with potentially operable intracranial lesions. Data of this nature can inform research, policy, and clinical collaborations that strengthen the neurosurgical capacity of low-income countries.**

INTRODUCTION

Hydrocephalus is a common neurosurgical disorder that affects people of all ages.[1] Hydrocephalus can result from any condition that interferes with cerebrospinal fluid dynamics, including congenital brain malformations, trauma, infection, neoplasm, and intracranial hemorrhage.[2] If not promptly diagnosed and treated, hydrocephalus can lead to significant disability or death. Stagno et al.[1] cite a prevalence of hydrocephalus in high-income countries ranging from 0.9 to 1.2 per 1000 population; however, little is known about hydrocephalus in low-income countries (LICs), where access to radiologic diagnosis often is lacking or absent. Whereas high-income countries have an estimated 42 computed tomography (CT) scanners per 1 million population, LICs have only 0.32 scanners per 1 million population.[3] Even where CT is available in LICs, it often is inaccessible or unaffordable for most patients.[3] Data on the

Key words
- Global neurosurgery
- Global surgery
- Haiti
- Hydrocephalus
- Neuroepidemiology

Abbreviations and Acronyms
CT: Computed tomography
ETV: Endoscopic third ventriculostomy
HUM: Hôpital Universitaire de Mirebalais
IVH: Intraventricular hemorrhage
LIC: Low-income country

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availability of magnetic resonance imaging scanners indicate that access to this technology is even more exclusive than for CT; for example, the regional density of magnetic resonance imaging scanners per 1 million population for the World Health Organization Africa region is 0.1, compared with a CT density of 0.4 per 1 million population for the same region.[4]

Limited data on pediatric hydrocephalus in LICs come from sub-Saharan Africa. With 36,000,000 births per year in sub-Saharan Africa, it is calculated that in this region, up to 180,000 new infant cases of hydrocephalus occur each year, and more than 100,000 newborns annually develop hydrocephalus before 1 year of age.[5,6] A disease-focused strategy of research and neurosurgical capacity building in Uganda has led to a deeper understanding of the burden of pediatric hydrocephalus in that country, where 60% of infant cases were reported to result from neonatal infection.[7] This work informed the development of innovative therapeutic approaches to address the disease burden in the Ugandan context, such as flexible endoscopic third ventriculostomy (ETV) with choroid plexus cauterization.[8]

Given evidence for effective strategies for the management of hydrocephalus in LICs, there is a need to better understand the burden and etiologies of hydrocephalus in this context for program and policy development. Here, we analyze radiologic patterns of hydrocephalus from a large sample of CT scans of the head performed at a public hospital in rural Haiti, an LIC in the Caribbean, examining causes of pediatric and adult hydrocephalus.

BACKGROUND

The Republic of Haiti occupies the western third of the Caribbean Island of Hispaniola. Its population is estimated to be 10.6 million as of July 2017, and its per capita GDP was estimated to remain unchanged at $1800 for the years 2015–2017.[9] With nearly 60% of the population living below the poverty line, Haiti is the, "poorest country in the Western Hemisphere." There are currently 4 neurosurgeons practicing in Haiti, all of who serve the population of the nation's urban capital, Port-au-Prince.[10] The current study was conducted at the Hôpital Universitaire de Mirebalais (HUM), a 205,000-square foot, 300-bed referral center in the rural city of Mirebalais in Haiti's Central Plateau.[11] HUM has a primary catchment of 185,000 patients and a tertiary catchment area of 3.4 million people from Mirebalais and surrounding regions.[11]

METHODS

Institutional Review Board Approval

Ethical approval for this study was obtained from the Institutional Review Boards of Zamni Lasante/Partners In Health, and Partners Healthcare.

Data Collection

CT studies of the head were obtained by local providers at HUM on a 32-slice helical CT scanner (BodyTom; Neurological/Samsung, Danvers, Massachusetts, USA), and stored locally on a picture archiving and communication system (Change Healthcare, Richmond, BC, Canada), mirrored in Boston, Massachusetts, USA. Studies were interpreted by 40 volunteer radiologists throughout North America. All radiologic reports were made available to clinicians in Haiti. Reports were deidentified, and data were analyzed using Microsoft Excel (Redmond, Washington, USA).

Data Analysis

The CT studies were performed between July 2013 and January 2016 at HUM, a public tertiary center in rural Haiti. Extracted data included demographics (age and sex of patient), referral source, study indication, and radiologic findings. CT reports were analyzed for findings consistent with hydrocephalus by evaluating radiology reports for the terms "hydrocephalus," "ventricular enlargement," trapped ventricles," "ventriculomegaly," and "enlargement of the ventricles." The reported etiology of hydrocephalus was determined from the radiologist's impression. Cases in which the radiologist proposed 2 or more possible etiologies without indicating which one was more likely were classified as ">1 possible etiology." Cases in which the radiologist could not differentiate between hydrocephalus and ex vacuo dilatation of the ventricles were classified as "atrophy present." In young children, although communicating hydrocephalus can be of either congenital or secondary etiology, no cases of communicating hydrocephalus in this data set were of exclusively congenital etiology. Among patients with noncommunicating hydrocephalus seen in this data set, the designation "congenital" was applied if: 1) the patient was <5 years and did not have an associated brain mass or intraventricular hemorrhage (IVH), or 2) if hydrocephalus was attributable to a congenital brain malformation, regardless of age.

RESULTS

During the study period, 3614 scans were performed on 3416 patients. Unless otherwise indicated, repeat scans were excluded from subsequent analyses. Of the 3416 initial scans, 119 (3.5%) had findings consistent with hydrocephalus. These 119 scans represented 6.3% of all initial scans with abnormal findings ($n = 1898$). Referrals for CT imaging of 88 (73.9%) of these patients originated from the emergency department, whereas the remaining patients came from the women's health ward (10 patients; 8.4%), the outpatient clinic (9 patients; 7.6%), the pediatrics ward (8 patients; 6.7%), the men's health ward (3 patients; 2.5%), and the neonatal intensive care unit (1 patient; 0.8%). The age of patients with hydrocephalus ranged from 0 to 90 years, with median age 35.5. **Table 1** shows the number of hydrocephalus cases observed in each age group. In total, 39% of cases occurred in pediatric patients (<18 years old) (**Table 1**); hydrocephalus was the second most common abnormal neurologic finding in pediatric patients after findings related to head trauma.

In 90% scans demonstrating hydrocephalus, the ordering clinician specified an indication for the head CT. **Table 2** shows the indications that led to discovery of hydrocephalus on imaging. Reported etiologies of hydrocephalus are shown in **Table 3**. Intracranial tumors were reported in 11 (55%) pediatric patients and 13 (29%) adults with noncommunicating hydrocephalus. IVH was reported in 24 adults older than 40 years old, accounting for 73% of noncommunicating hydrocephalus in that age group (see **Table 3** for detailed breakdown). Excluding 7 cases in this cohort for which the ordering clinician provided a history of trauma (**Table 2**), most cases of IVH and other intracranial hemorrhage in this series

Table 1. Proportion of Hydrocephalus Among Haitian Patients Undergoing Head CT by Age Group

Age Group	Number of Abnormal Scans	Number of Scans with Hydrocephalus (% of Total Abnormal)	Hydrocephalus in Age Group as % of Total Scans with Hydrocephalus
Children	261	46 (18%)	38.6%
<1 year	32	9 (28%)	7.6
1–4 years	70	16 (23%)	13.4
5–17 years	159	21 (13%)	17.6
Adults	1635	73 (4%)	61.4
18–39 years	573	21 (4%)	17.6
40–59 years	558	23 (4%)	19.3
60+ years	504	29 (6%)	24.4
Age not reported	2	0	0
Grand total	1898	119 (6%)	100

Percentages in fourth column represent percentage of abnormal scans in that age group that had findings consistent with hydrocephalus.
CT, computed tomography.

Table 2. Indications Listed by Ordering Clinicians in Scans with Findings of Hydrocephalus

Indication	Number (%) of Scans
Suspected stroke	32 (27%)
Motor deficit	13 (11%)
Seizure or epilepsy	12 (10%)
Mental status change	12 (10%)
No indication listed	12 (10%)
Oncologic indication	8 (7%)
Known hydrocephalus	8 (7%)
Language issue or aphasia	7 (6%)
Trauma	7 (6%)
Headache	5 (4%)
Coma	5 (4%)
Focal deficit NOS	4 (3%)
Macrocephaly	3 (3%)
Suspected infection	3 (3%)
Loss of consciousness	2 (2%)
Developmental delay	2 (2%)
Shunt malfunction	1 (1%)
Meningomyelocele	1 (1%)

Scans in table add up to more than 119, as each scan could have more than one indication.
NOS, not otherwise specified.

were hemorrhagic strokes, an important cause of morbidity and mortality as described in other reports on Haitian cerebrovascular epidemiology.[3,10,12-14] In contrast to the patients older than 40 years of age in this sample, only a single case of IVH was identified among pediatric patients with hydrocephalus, and 2 additional cases were found in the 18–39 age group (Table 2). Of the 119 patients examined in this study, 10 had follow-up CT scans within the study period. On first follow-up scan, 2 patients had reported interval worsening of their hydrocephalus and 3 had no significant changes. For the remaining 5 patients, the radiologists did not comment on the evolution of the hydrocephalus.

DISCUSSION

In this retrospective study using head CT data from a public hospital in rural Haiti, 3.5% of patients had imaging consistent with hydrocephalus, a neurosurgical disease. Indeed, shunting for hydrocephalus is 1 of only 2 neurosurgical conditions listed in the World Bank's list of essential surgical procedures.[15] Patients in this study with hydrocephalus ranged from 0 to 90 years of age, the mean age was 35.5, and pediatric cases (<18 years old) represented 39% of cases (Table 1). Among all hydrocephalus cases in this cohort, 29% were found to have communicating hydrocephalus without evidence of associated subarachnoid or IVH, whereas 55% of these patients had radiological evidence of non-communicating hydrocephalus (Table 3).

Intracranial tumors were a common cause of obstructive hydrocephalus in this study cohort (Table 3), identified in 55% of pediatric patients and 29% of adults with noncommunicating hydrocephalus. Data on the epidemiology of brain tumors in LIC are limited due to limited availability of diagnostic neuroimaging and neurosurgical care.[3,16-18] A retrospective operative series from Uganda found that symptomatic hydrocephalus was the most common presentation among pediatric patients with brain tumors, accounting for 66.9% of the study cohort; among 41.3% of patients in that study, presentation was "visible," such as macrocephaly or a visible mass.[18] Our analysis of adult hydrocephalus cases in Haiti provides insights into etiologies of adult hydrocephalus in LIC, another entity for which little data from LIC are available.[1,19] Noncommunicating hydrocephalus predominated in patients aged 40–60 years old, with brain tumors and IVH accounting for the majority of cases in this age group (Table 3). In adults >40 years of age, 73% of noncommunicating hydrocephalus was due to IVH. IVH is a condition in which outcomes are likely to be improved with cerebrospinal fluid diversion, with mortality estimates ranging from 50% to 80% without this intervention.[16,20,21] Our data therefore underscore the importance of developing neurosurgical capacity for adults with hydrocephalus in rural LICs.

Our findings contribute to a growing body of literature on the burden of hydrocephalus in low- and middle-income countries. A recent systematic review examining the global epidemiology of pediatric hydrocephalus found that the pooled incidence of this disease entity among World Health Organization regions was greatest among Latin American countries, at 316 per 100,000 births. Estimates in the African region were 145 per 100,000

Table 3. Likely Cause of Hydrocephalus Classified by Age Group

	Age Group, years						
	<1	1–4	5–17	18–39	40–59	60+ years	Total
Communicating	3 (33%)	8 (50%)	5 (24%)	8 (38%)	4 (17%)	8 (28%)	36 (30%)
SAH	0 (0%)	0 (0%)	0 (0%)	1 (5%)	1 (4%)	1 (3%)	3 (3%)
>1 possible etiologies	3 (33%)	8 (50%)	5 (24%)	7 (33%)	3 (13%)	7 (24%)	33 (28%)
Noncommunicating	3 (33%)	5 (31%)	13 (62%)	12 (57%)	19 (83%)	14 (48%)	66 (55%)
Congenital*	2 (22%)	1 (6%)	1 (5%)	0 (0%)	0 (0%)	0 (0%)	4 (3%)
IVH	0 (0%)	0 (0%)	1 (5%)	2 (10%)	11 (48%)	13 (45%)	27 (23%)
Tumor	0 (0%)	1 (6%)	10 (48%)	7 (33%)	5 (22%)	1 (3%)	24 (20%)
>1 possible etiologies	1 (11%)	3 (19%)	1 (5%)	3 (14%)	3 (13%)	0 (0%)	11 (9%)
Type not specified	3 (33%)	3 (19%)	3 (14%)	1 (5%)	0 (0%)	1 (3%)	11 (9%)
Atrophy present	0 (0%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)	6 (21%)	6 (5%)
Total	9	16	21	21	23	29	119

Percentages represent percentage of scans in each age group that specified a particular cause of hydrocephalus.
SAH, subarachnoid hemorrhage; IVH, intraventricular hemorrhage.
*In this study, congenital were only seen among pediatric patients (<18 years) with noncommunicating hydrocephalus that was not attributable to a brain mass or IVH. The 4 cases included: a case of "likely aqueductal stenosis," a case of "Blake pouch cyst/Dandy Walker continuum," a case of Chiari malformation, and a case of "vein of Galen malformation."

births, whereas the lowest estimates were found in North America, where incidence was estimated at 68 per 100,000 births.[19]

This study has limitations. Analysis of CT reports in isolation does not provide the full clinical context of these patients. Some etiologies of hydrocephalus cannot be determined by imaging data alone (e.g., without access to serological and/or cerebrospinal fluid studies to evaluate for infection), and patient outcomes are unknown. CT data were obtained from a tertiary referral center, which may have caused referral bias that could enrich our sample with more ill and/or complex patients.

CONCLUSIONS

Our findings are notable for a series of patients in a rural LIC setting with CT evidence of hydrocephalus, a neurosurgically treatable disorder. Most patients presented with noncommunicating hydrocephalus for which the most common associated findings were IVH and brain tumors. Although patients spanned all age groups, there was a predominance of children and young adults with this diagnosis, and associated findings varied by age; IVH, for example was observed with greater frequency among adults than among pediatric patients in this cohort.

Neurosurgical intervention for hydrocephalus includes 2 common approaches to cerebrospinal fluid diversion: ventriculoperitoneal shunting or ETV; in patients younger than 2 years of age, ETV may be performed in conjunction with choroid plexus cauterization. The noninferiority of the latter approach to the former in regard to developmental outcome and failure rate at 1 year for Ugandan infants <6 months of age with postinfectious hydrocephalus was demonstrated in a recent single-center randomized trial.[22] As in many other LICs, the country of Haiti currently has an exceedingly small number of clinical neuroscience providers: 4 neurosurgeons and 2 neurologists, for a population of 10.6 million people.[10,23-27]

REFERENCES

1. Stagno V, Navarrete EA, Mirone G, Esposito F. Management of hydrocephalus around the world. *World Neurosurg*. 2013;79(2 suppl). S23.e17-20.
2. Kestle JRW, Kulkarni AV, Warf BC. Introduction: Pediatric hydrocephalus: a continuing evolution in our understanding and management. *Neurosurg Focus*. 2016;41:E1.
3. Valtis YK, Cochran MF, Martineau L, Lamour B, Mendel JB, Berkowitz AL. Head CT findings at a public hospital in rural Haiti. *J Neurol Sci*. 2017;379:327-330.
4. World Health Organization. *Global atlas of medical devices - 2014*. Geneva: World Health Organization; 2014.
5. Muir RT, Wang S, Warf BC. Global surgery for pediatric hydrocephalus in the developing world: a review of the history, challenges, and future directions. *Neurosurg Focus*. 2016;41:E11.
6. Warf BC. Educate one to save a few. educate a few to save many. *World Neurosurg*. 2013;79(2 suppl). S15.e15-8.
7. Warf BC, Dagi AR, Kaaya BN, Schiff SJ. Five-year survival and outcome of treatment for postinfectious hydrocephalus in Ugandan infants. *J Neurosurg Pediatr*. 2011;8:502-508.
8. Warf BC. "Who is my neighbor?" Global neurosurgery in a non-zero-sum world. *World Neurosurg*. 2015;84:1547-1549.
9. Central Intelligence Agency. The World Factbook—Central Intelligence Agency. Available at: https://www.cia.gov/library/publications/resources/the-world-factbook/geos/ha.html. Accessed November 10, 2017.
10. Barthélemy EJ, Benjamin E, Edouard Jean-Pierre MY, Poitevien G, Ernst S, Osborn I, et al. A prospective emergency department-based study of pattern and outcome of neurologic and

neurosurgical diseases in Haiti. *World Neurosurg*. 2014;82:948-953.

11. Partners in Health: Hôpital Universitaire de Mirebalais | Partners In Health. Available at: https://www.pih.org/pages/mirebalais. Accessed July 26, 2018.

12. Kenerson JG. Hypertension in Haiti: the challenge of best possible practice. *J Clin Hypertens*. 2014;16:107-114.

13. Koch S, Pabon D, Rabinstein AA, Chirinos J, Romano JG, Forteza A. Stroke etiology among Haitians living in Miami. *Neuroepidemiology*. 2005;25:192-195.

14. Lavados PM, Hennis AJ, Fernandes JG, Medina MT, Legetic B, Hoppe A, et al. Stroke epidemiology, prevention, and management strategies at a regional level: Latin America and the Caribbean. *Lancet Neurol*. 2007;6:362-372.

15. Debas HT, Donkor P, Gawande A, Jamison DT, Kruk ME, Mock CN. *Disease Control Priorities*. In: *Essential Surgery*. 3rd ed. Vol. 1. Washington, DC: World Bank; 2015.

16. Hanley DF. Intraventricular hemorrhage severity factor and treatment target in spontaneous intracerebral hemorrhage. *Stroke*. 2009;40:1533-1538.

17. Mclane HC, Berkowitz AL, Patenaude BN, Mckenzie ED, Wolper E, Wahlster S, et al. Availability, accessibility, and affordability of neurodiagnostic tests in 37 countries. *Neurology*. 2015;85:1614-1622.

18. Stagno V, Mugamba J, Ssenyonga P, Kaaya BN, Warf BC. Presentation, pathology, and treatment outcome of brain tumors in 172 consecutive children at CURE Children's Hospital of Uganda. The predominance of the visible diagnosis and the uncertainties of epidemiology in sub-Saharan Africa. *Child's Nerv Syst*. 2014;30:137-146.

19. Dewan M, Rattani A, Mekary R, Glancz L, Yunusa I, Baticulon R, et al. Global hydrocephalus epidemiology and incidence: systematic review and meta-analysis [e-pub ahead of print]. *J Neurosurg*. https://doi.org/10.3171/2017.10.JNS17439, accesed January 12, 2018.

20. Hinson HE, Hanley DF, Ziai WC. Management of intraventricular hemorrhage. *Curr Neurol Neurosci Rep*. 2010;10:73-82.

21. Weinstein R, Ess K, Sirdar B, Song S, Cutting S. Primary intraventricular hemorrhage: clinical characteristics and outcomes. *J Stroke Cerebrovasc Dis*. 2017;26:995-999.

22. Kulkarni AV, Schiff SJ, Mbabazi-Kabachelor E, Mugamba J, Ssenyonga P, Donnelly R, et al. Endoscopic treatment versus shunting for infant hydrocephalus in Uganda. *N Engl J Med*. 2017;377:2456-2464.

23. Berkowitz AL. All for one. JAMA. 2015;314:1341.

24. Berkowitz AL, Martineau L, Morse ME, Israel K. Development of a neurology rotation for internal medicine residents in Haiti. *J Neurol Sci*. 2016;360:158-160.

25. Park KB, Johnson WD, Dempsey RJ. Global neurosurgery: the unmet need. *World Neurosurg*. 2016;88:32-35.

26. Punchak M, Mukhopadhyay S, Sachdev S, Hung Y-C, Peeters S, Rattani A, et al. Neurosurgical care: availability and access in low- and middle-income countries. *World Neurosurg*. 2018;112:e240-e254.

27. World Federation of Neurosurgical Societies: Global Neurosurgical Workforce Map - About WFNS | WFNS. World Federation of Neurosurgical Societies. Available at: https://www.wfns.org/menu/61/global-neurosurgical-workforce-map. Accessed October 27, 2017.

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