Barriers to Spina Bifida Care in Benin: A Single-Center Hospital-Based Cross-Sectional Study

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Abstract:

Introduction

Spina bifida (SB) is one of the common birth defects of the spinal cord and leads to permanent disability for newborns and young children. The management of this condition has faced barriers in the Republic of Benin. We aimed to assess the current prevalence of SB, management, in Benin, as well as the availability of pediatric neurosurgery in Benin.

Methods

SB patients admitted at the authors' institution between January 2018 and August 2021 were included. Demographics, access to care, and therapeutic data were extracted from patient records.

Results

Forty patients aged 99 days (95% CI:5.9-192.1) were included. Most were male (24, 60%), 23 (57.5%) experienced a delay in diagnosis after birth ranged from 5 days - 7months and 28 (70%) could not afford neuroimaging. Most lesions were lumbosacral (n=23, 57.5%). Five patients had comorbidities, the most prevalent of which was club foot (4, 10%). Twenty-two patients (55%) had surgery repair, and the admission-to-surgery delay ranged from 2 days - 2 years. Sixteen patients (40%) required a ventriculoperitoneal shunt, and they were more likely to experience delays (OR=11.67, 95% CI:2.14-63.64, p=0.02). Three patients died (7.5%), and one developed meningitis (2.5%).

Conclusion

SB care in Benin remains suboptimal due to the lack of access to care, and cultural barriers like the belief in a curse of the gods, or infidelity of the woman.

The delay between care and the diagnosis is long because of the unavailability of time and cost of neuro-imaging, cost of the device, and consumables for surgery.

KEYWORDS: Spina bifida, Benin, Global neurosurgery, Hydrocephalus, Pediatric neurosurgery.

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Introduction

Spina Bifida (SB) is a serious birth defect resulting from improper neural tube closure after approximately 4 weeks of gestation, occurring in 1-10 per 1,000 live births worldwide and leading to permanent disability.[1-4] The burden of SB is high in Africa, with a pooled multinational continental prevalence of 0.13%, but is scarcely reported, given this condition is not prioritized in national health planning.[5-7] Although the population prevalence of spina bifida in Benin has not been reported, the hospital prevalence is 0.057%, with delay in treatment due to delayed or non-existent antenatal diagnosis, due to financial challenges cultural barriers.[8]

The causes of SB are multifactorial, but insufficient maternal dietary folic acid intake is known to be a prime cause.[1-9] No family history is present in 90% of parents of infants affected with SB.[10] Established risk factors include the history of previously affected pregnancy with the same partner, pregestational maternal diabetes, and use of valproic acid or carbamazepine.[11] Suspected risk factors include maternal Vitamin B12 deficiency, maternal obesity, maternal hyperthermia, and maternal diarrhea, among others.[11] However, ensuring adequate intake of folic acid (Vitamin B9) among pregnant women prevents approximately 70% of cases of SB.[6-12] Folate fortification policies have been particularly effective in reducing the incidence of SB.[7]

In low- and middle-income countries (LMICs), including Benin, the burden of pediatric neurosurgical disease continues to grow due to inadequate pediatric neurosurgical workforce density, the lack of equipment, a paucity of funding, and poor healthcare infrastructure.[13-14] Children with SB often do not receive necessary surgical intervention. In this study, we aimed to assess the current prevalence of SB, management, and rehabilitation practices in Benin, as well as the availability of pediatric neurosurgery in Benin.

Methods

Ethical approval

Ethical approval was not required because only de-identified data were collected. Patient consent was neither required nor sought for the same reason.

Setting

The authors conducted a cross-sectional study with retrospective data collection at a Beninese neurosurgery referral center, Teaching Hospital Hubert K Maga of Cotonou.

Data collection and extraction

The authors accessed the records of spina bifida patients admitted at the aforementioned institution between January 2018 and August 2021. The extracted data included: demographics (i.e., age, sex), access to care data (determinants of delays in seeking, reaching, and receiving care), neuroimaging, treatment received (surgical or non-surgical), and patient outcome. Incomplete or missing patient data were treated first by triangulation, in which other forms of data collection, such as patient interviews, were used to complement chart review data. Patient data that were incomplete despite triangulation were excluded.

Statistical analysis

Summary descriptive statistics were computed for parametric (i.e. mean and 95% confidence intervals [CI]) and non-parametric (i.e., median and interquartile range [IQR]) data. Bivariable associations were calculated using the Fisher's Exact and Mann-Whitney U tests as appropriate. Also, odds ratios were collected for select variables. Results were considered significant when the p-value was <0.05.

Results

Forty patients with SB were admitted between 2018 and 2021 with a peak in 2020 (n=15, 37.5%) (Figure 1). There were more male patients than female patients overall (n=24, 60% vs. n=16, 40%) and the male prevalence was higher each year, except in 2021 (p=0.31). The mean age at diagnosis was 99 days (95% CI: 5.9-192.1) and male patients were older than female patients (111.1 vs. 80.8 days, p=0.45). The majority of mothers received antenatal care from a skilled health professional (n=24, 60%), but a minority received folic acid supplementation (n=11, 27.5%).

The diagnosis was made clinically and radiologically. Twenty-three (57.5%) patients experienced a delay in diagnosis after birth ranged from 5 days – 7 months, and 28 (70%) could not afford neuroimaging. SB lesions were lumbosacral (n=23, 57.5%), lumbar (n=13, 32.5%), sacral (n=2, 5%), dorsolumbar (n=1, 2.5%), and sacrococcygeal (n=1, 2.5%). Twenty-one (52.5%) of these lesions were ulcerated. Five patients had comorbidities including club feet (n=4, 10%) and corpus callosum agenesis (n=1, 2.5%).

All patients were offered surgical repair. Twenty-two patients (55%) had surgery, but 18 (45%) could not afford surgery, so surgery was offered *pro bono* by surgical mission ships with the surgical team at the reference hospital HKM of Cotonou in Benin. The admission-to-surgery delay ranged from 2 days to 2 years, with many patients (n=10, 25%) being operated on within a month of admission. A ventriculoperitoneal shunt was placed in 16 cases (40%). These patients were more likely to experience delays (OR=11.67, 95% CI:2.14-63.64, p=0.02) (Table 1).

However, six (15%) patients declined care and left the hospital against medical advice. Of note, two patients required redo surgery (5%) of patients who benefited from surgery. Three patients (7.5%) died, one (2.5%) had meningitis, and 17 (42.5%) were lost to follow-up.

Discussion

We explored the landscape of SB at a Beninese neurosurgical referral center. We found more male than female patients and significant barriers to care. Lumbosacral SB was more prevalent, and most of these cases were ulcerated. Patients faced barriers at every step of the continuum of care, including financial barriers. Only 27.5% of mothers received folic acid supplementation to prevent SB. However, it will be mandatory that the folate must be given before conception.

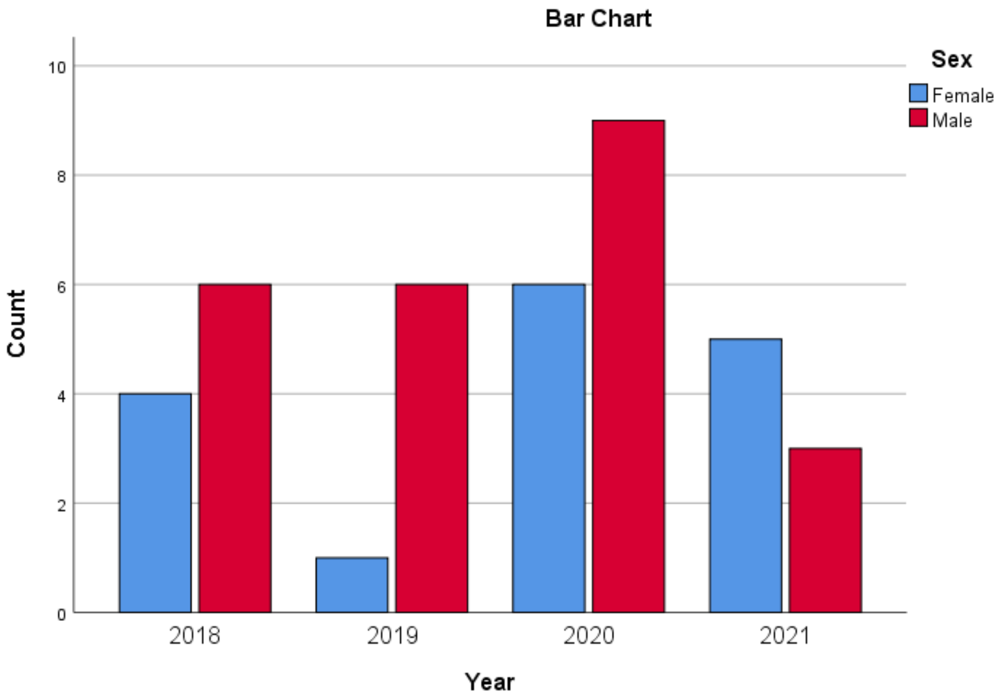

Figure 1. Annual distribution of spina bifida patients in Benin disaggregated by sex

Variables	Odds ratio	95% CI	P-value
Sex (Female/Male)	1.67	0.46-6.01	0.52
Two vertebral levels involved	1.31	0.36-4.78	0.75
Antenatal care	2.57	0.70-9.46	0.20
Could not afford neuroimaging	2.52	0.63-10.05	0.30
Could not afford surgery	1.67	0.46-6.01	0.52
Surgical repair	0.64	0.18-2.28	0.54
Ventriculoperitoneal shunt	11.67	2.14-63.64	0.02*

Table 1. Factors associated with delays in seeking, reaching, and receiving care among Beninese spina bifida patients

The cases of neural tube defects occurring per year are approximately between 35.622 to 300000 globally, causing significant mortality and morbidity.[15-16] Africa bears more than 64% of the global neural tube defect disease burden.[15] Our hospital-based study identified 40 cases between January 2018 and August 2021. These results are likely an underestimation considering that Benin's birth rate is 41.6 births per 1,000 population, and congenital birth defects account for 7.2% (95%CI: 3.4-11.9%) of disease burden in the under-five age group.[17] This supposition is supported by the high proportion of barriers to seeking, reaching, and receiving care in our cohort. Further population-based epidemiological studies are necessary to quantify the birth prevalence of SB in Benin.

The People and Organizations United for Spina Bifida and Hydrocephalus' (PUSH!) 2016 spina bifida scorecard rates Benin at 2.5 stars.[18] The scorecards report on four items: data availability, implementation of folic acid fortification and supplementation policies, access to care, and quality of care.19 Scores between 0 and 3 indicate a need for improvement, scores of 4 or 5 are good, and a score of 6 is excellent.[19] Benin's 2.5 stars are an aggregate of prevention (1 star), access to care (0.5 star), and quality of life (1 star).[18] The one star for prevention indicates Benin has implemented mandatory folic acid fortification of wheat flour, though consumption of wheat flour may be inadequate, particularly in rural Benin.[18-20] However to this day no study was done in Benin to show that the rate of SB changed after the folate requirement. The one-star quality of life rating indicates Benin has signed and ratified the United Nations Convention on the Rights of Persons with Disabilities.[18] Benin's 0.5 star access to care rating reflects an inadequate neurosurgical workforce density between 0.2 and 1 per 1 million people.[18]

Our study likely underreports the birth prevalence of SB in Benin. Skilled birth attendants should be trained to detect neural tube defects, and a national congenital diseases registry should be maintained to monitor the birth prevalence of SB and other congenital malformations. The congenital disease registry reporting mechanism may be based on existing successful reporting models such as the human immunodeficiency virus. In addition, the use of folic acid supplements was low in our study. A previous study indicated that a policy approach consisting of mandatory food fortification with folic acid and folic acid supplementation is optimal in reducing the birth prevalence of folate-preventable SB.[7] Neurosurgeons are building a multispecialty group of stakeholders, with a plan to introduce a resolution for folate fortification at the World Health Assembly. Public health campaigns aimed at increasing awareness regarding SB and the role of folic acid in its pathogenesis are necessary to increase periconceptional intake of folic acid. A level of 400 ugs of folic acid per day in the form of supplements has been utilized previously in Benin, in line with standard recommendations for the prevention of SB.[18-20]

The proportion of mothers who did not receive antenatal care and age at diagnosis are major causes of concern. These factors contribute to delayed care and avoidable morbidity. Beninese SB patients faced significant financial risk, leading to delays in receiving care. For this reason, it is essential that SB patients be covered by national health insurance schemes, particularly because the national poverty rate exceeds 38.5% (rural: 39.7% and urban: 31.4%).[17] The fight for the financial risk protection of Beninese SB patients should prompt collaboration between neurosurgeons, orthopedic surgeons, urologists, geneticists, and patient advocacy groups. Importantly, a broad coalition of stakeholders should interface with policymakers to affect policy change aimed at reducing the birth prevalence of SB and increasing access to care for children with SB.

Conclusion

The use of folic acid supplements among mothers of children with SB in Benin is low. SB care in Benin is often delayed due to the financial burden on families. Population-based registries are necessary to accurately ascertain the burden of SB in Benin, while public awareness campaigns are required to increase awareness of the utility of folic acid supplements in preventing SB. Importantly, policy-level action is necessary to scale up neurosurgical capacity for treating SB patients in Benin.

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