ORIGINAL ARTICLE

Managing Children with Spina Bifida in sub-Saharan Africa: *the Zambian experience?*

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ABSTRACT

Purpose: To investigate the management of children with SB and outcome measures used in Zambia.

Methods: A retrospective cross-sectional study was done. Between 2001 and 2010, a total number of 253 children with SB who were managed at both the University Teaching Hospital (UTH) and Beit Cure Hospital (BCH) were identified.

Results: Majority (56%) of the patients were aged between 1-6 months ($p < 0.001$). Hydrocephalus was prevalent in 61% of the patients. Myelomeningocele was the most common (61%) defect and the lumbar region was the common site (60%) ($p < 0.001$). Majority (28%) of children came from the Southern Province of Zambia ($p < 0.001$). The majority (81%) of patients were lost to follow-up ($p < 0.001$). None of the files had outcomes measuring instruments.

Conclusions: There is evidence that interventions were given although the outcomes were not measured. The majority of the children came from the Southern Province of the country. A study ought to be done to investigate the predominance of the prevalence of SB in that part of Zambia. Many patients were lost to lack of follow-up, hence proper follow-up mechanisms must be instituted by both hospitals. Researchers are challenged to develop measuring instruments that are culturally sensitive and appropriate to the needs of Zambian children.

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INTRODUCTION

Managing children with Spina Bifida (SB) in sub-Saharan Africa can be potentially devastating due to prevailing low socioeconomic status, harmful taboos, religious beliefs, paucity of medical power and inadequate facilities[1-5]. Spina bifida is considered the commonest of the neural tube defects (NTDs) and hydrocephalus commonly occurs in association[6-9]. Notwithstanding the striking down in the incidence of NTDs over the past three decades, Zambia is reported to have high incidence rates in Africa despite the global decline[10]. It is one of the few African countries that have a policy on folic acid food fortification, but prevention of NTDs which are life-long disabilities require a good diet of fruits and vegitables or supplements for all child bearing women[11].

Spina bifida and hydrocephalus are the most recurrent and disabling malformations in neonates in the sub-African pediatric environment. Studies done in Nigeria, Cameroon, Kenya, Uganda on

Key words: *hydrocephalus, ventriculoperitoneal shunt, endoscopic third ventriculostomy, choroid plexus cauterization, physiotherapy, HIV/AIDs counselling*

management of children with SB have reported challenges encountered in the management[1-4]. However, long-term outcomes of management and outcome measures used to evaluate the impact of interventions given to children with SB in the sub-Saharan Africa region are not well known.

In the recent past, there has been increasing emphasis on evaluation of therapy outcomes by rehabilitation professionals worldwide. Literature has highlighted some common questions that are asked by clinicians concerning their interventions and some of these questions are as follows: *Am I an effective clinician? How many of my patients achieve their goals? Are my patients satisfied with their gains? And can my treatments be more efficient or effective*[12]*? Of particular importance is the ability to note the need to move beyond efficacy and effectiveness to consider the health economics of rehabilitation programs if they have to be relevant in the real world of provision of health interventions*[13].

Given the concepts associated with rehabilitation and challenges encountered in executing interventions of rehabilitation for children with SB especially in Africa, some of the key issues that need to be addressed are '*what works, given the nature of facilities available*?', '*how can that which works be measured?', 'what can be used to measure what works*?' The issue of what defines a '*good*' outcome remains a question of major focus in research, clinical practice and policy[14]. In essence, the International Classification of Function, disability and health (ICF) that focuses on the functioning aspect of an individual looks at outcomes with the practice-based evidence (PBE) research design, which focuses on process and also quantifies person and outcomes details[15,16].

Considering the intricacy of SB, the far reaching severity, the demand for sustained and often invasive care and the implication for the child and the family, it is salient that the evidence base of the current care and outcomes of the management in Zambia is very thin. The relative scarcity of studies on the rehabilitation outcomes of children with SB in Zambia is very striking. Considering such limitations, we set out to evaluate the management and the outcome instrument measures used in measuring the impact of rehabilitation in children with SB in Zambia.

METHODOLOGY

A descriptive retrospective cross-sectional research design with quantitative methods was utilised. Ethical approval was obtained from the Biomedical Research Ethics Committee of the University of Zambia. This article is part of the first author's PhD study in the Department of Physiotherapy, at the School of Medicine, University of Zambia.

The research study was conducted under the auspices of the UTH and BCH. Both hospitals are found in Lusaka and are the only centres providing specialized care to children with SB and hydrocephalus in Zambia. The UTH is largest referral hospital in the country that provides several services to the general public by providing specialized diagnostic and surgical procedures, and also conducts research on commonly occurring diseases in the country. The BCH has been in operation since 2007 and provides specialized care to children with spina bifida (SB) and hydrocephalus. It is a private hospital that provides surgical facilities to children with physical disabilities.

Recruitment of participants and data capturing

A total of 253 files of children with SB were identified from the files of children with neurological problems who were were managed at both the UTH and BCH from January 2001 to January 2010. Demographic and patient characteristics of the children with regards to the variables of gender, age, provinces of origin and referral, type and location of SB, interventions given and the period of treatment were investigated using a checklist.

Age range at time of surgical repair was categorized into five groups: 0-3 weeks, 1-6 months, 7-11 months, 1-5 years and 6-10 years. The province of

origin and referral was taken note of. Type of SB was clustered into three groups: myelomeningocele, meningocele and lipomeningocele. The site of defect was categorized into seven groups: cervical, thoracic, thoracolumbar, lumbar, lumbosacral, sacral and sacral-coccygeal.

The surgical interventions given were grouped into ventriculoperitoneal shunt (VPS), endoscopic third ventriculostomy (ETV), choroid plexus cauterization (CPC), Z-Plasty, Ponseti and other procedures. Requests for physiotherapy were checked for and the central nervous system (CNS) assessments were investigated in order to verify the need for physiotherapy. The management period was censored from the first day of admission to the last review. Lastly, the presence of outcome instrument measures for evaluating the interventions given was investigated.

Data analysis

Descriptive statistics was utilized to analyze the data using SPSS version 17. Categorical variables were compared using either the Fishers' Exact test or Chi-square test. Censoring of treatment time was estimated using Kaplan-Meier methods. The level of statistical significance was set at $p \leq 0.05$ *at 95%* Confidence Interval.

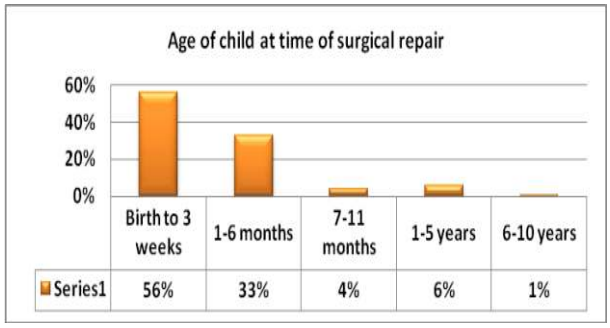
RESULTS

Demographic characteristics of the children with SB

Gender of child and age at the time of repair

Results showed that there were 139 (55%) males and 114 (45%) females ($p = 0.116$). A total of 143 (56%) children who underwent repair, were between the age range of 0-3 weeks, 83 (33%) were between 1-6 months, 9 (4%) between 7-11 months, 15 (6%) between 1-5 years and the lowest were 3 (1%) who represented the category of the age range of 6-10years ($p < 0.0001$).

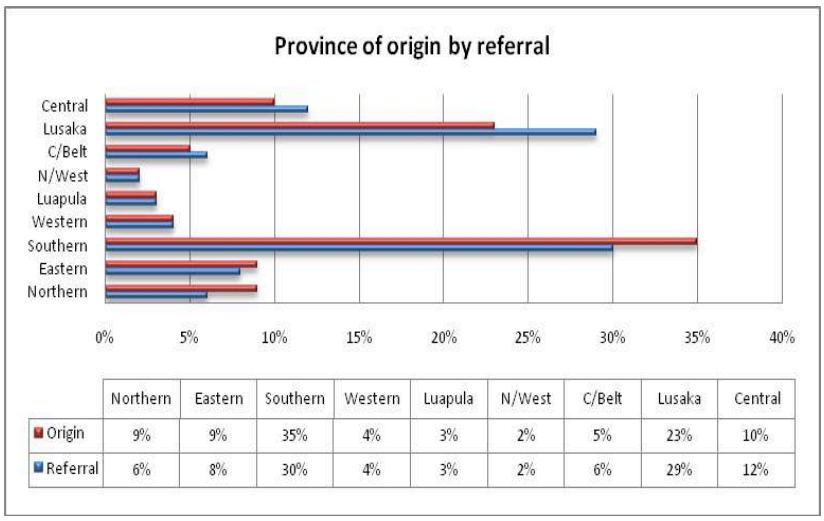
Figure 1: Age of child at time of surgical repair



Province of origin and referral

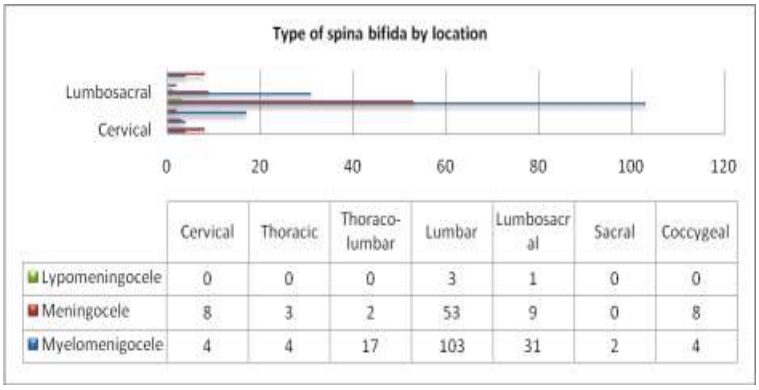
It was noted that 88 (35%) children originally came from the Southern Province of the country followed by Lusaka Province with 58 (23%) and the lowest province represented was the North-Western Province with 7 (3%) ($p < 0.0001$). The pattern of province of origin was the same with regard to province of referral where 77 (30%) children were referred from the Southern Province of the country followed by Lusaka Province with 74 (29%) and the lowest province represented was the North-Western Province with 5 (2%) ($p < 0.0001$).

Figure 2: Province of origin by referralPresence of hydrocephalus, type and location of Spina bifida



It was eminently seen that from all the files identified, a total of 155 (61%) patients were diagnosed with hydrocephalus while 98 (39%) had SB alone ($p < 0.0001$). The overall results (figure 2) showed that from the 253 children with SB, 154 (61%) had myelomeningocele, 79 (31%) meningocele and 4 (2%) lipomenigocele ($p < 0.0001$). Additional results with regards the site of the defect showed that, the lumbar region had 143 (60%), lumbo-sacral 41 (17%), thoracolumbar 19 (8%), sacral 13 (6%), cervical 12 (5%), thoracic 7 (3%) and lastly sacral-coccygeal 2 (1%) ($p < 0.0001$).

Figure 3: Type of spina bifida by location



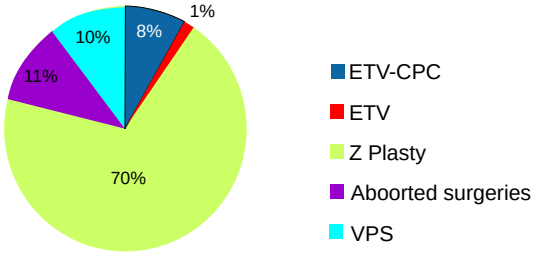
	Cervical	Thoracic	Thoraco-lumbar	Lumbar	Lumbosacral	Sacral	Coccygeal
Lypomeningocele	0	0	0	3	1	0	0
Meningocele	8	3	2	53	9	0	8
Myelomenigocele	4	4	17	103	31	2	4

Interventions given to the children

Surgical Interventions

The results showed that 177 (70%) underwent repair using the Z-plasty, 25 (10%) received VPS & repairs, 21 (8%) the procedure of ETV-CPC & repairs, and 3 (1%) went for ETV & repairs. Nonetheless, 27 (11%) were aborted surgeries due to various logistical problems ($p < 0.0001$).

Figure 4: Surgical Interventions



However, the results also showed that a few days after surgery, 142 (56%) children had reported clean wounds, 60 (24%) had infected wounds, 24 (9%) leaking CSF and 27 (11%) were aborted surgeries (($p < 0.0001$).

Physiotherapy and other forms of management

From the CNS assessments investigated, only 188 (74%) files had assessments done while, in 65 (26%) files, assessment forms were not seen. However, from those assessed, 116 (62%) had reduced tone, 42 (22%) had increased tone and 30 (16%) had normal tone. With regards physiotherapy requests, the service was poorly utilized where only 26 (24%) were referred and 90 (76%) were not referred from those with reduced tone while 10 (24%) were referred from those with increased tone and 32 (76%) were not referred ($p < 0.0001$).

Figure 5: Physiotherapy requests

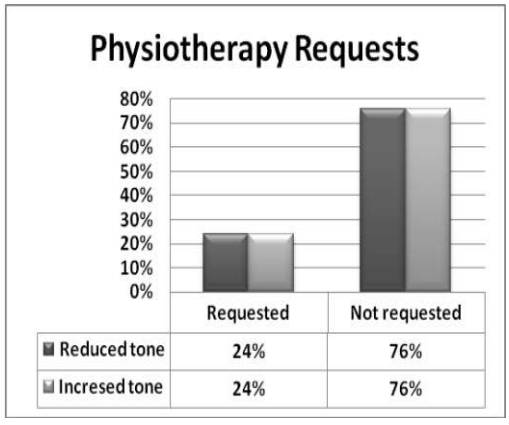
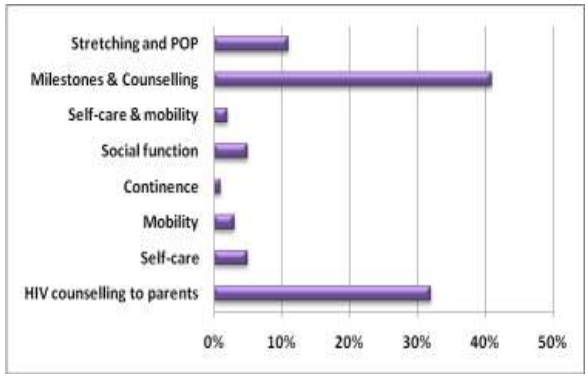


Figure 6: Other forms of management considered

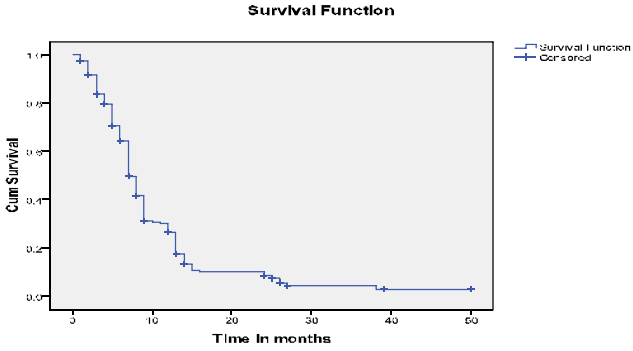


Results of the study also showed that HIV counselling (32%) to parents was given an important place together with delayed milestones and counseling (41%). Other issues considered were self-care, mobility, social function, continence and stretching and POP casting for foot correction.

Compliance to the general management

Generally, there was non-compliance in the patients where 206 (81%) were not so regular with management and only 47 (19%) were compliant ($p < 0.0001$). Additionally, 193 (76%) were only seen in a period of less than one year, 45 (18%) were seen in about one year, 12 (5%) in 2 years, 2 (0.8%) in 3 years and 1 (0.2%) was the only child seen for four years ($p < 0.0001$). Management time was censored using the Kaplan Meier (Figure 7).

Figure 7: Treatment time censored to show compliance in management



Instruments used for measuring outcomes of interventions

It was noted that none of the 253 files of children with SB had any kind of instrument measure that was used to evaluate the impact of interventions, except for the standardised assessment forms for serial head circumference measurements in those children that had hydrocephalus.

DISCUSSION

Managing children with spina bifida (SB) and hydrocephalus in sub-Saharan Africa can be potentially distressing due to several socioeconomic and health care associated factors. As observed in this study, managing 253 children with SB was quite overwhelming. Primary management was provided for, but a great number of the children were lost to follow-up. It has also been observed that physiotherapy services were quite under utilized and yet with a multidisciplinary approach, a great number of children being managed can be followed up if referred to local physiotherapy departments. This will bring about stability in the children hence promoting meaningful outcomes in order to avoid delayed milestones and physical disabilities.

Demographic and clinical characteristics of the patients

It has been observed that sex differences clearly exist in the prevalence of SB. It is intriguing to note in literature that at birth, SB tends to be more common in girls than in boys at a sex ratio of 1/0.8 obtained from a study that showed that 70% of the neonates were girls while 30% were boys[17-19]. Other studies are also consistent with findings that females are more likely to have SB than males and this predominance appears to be influenced by the presence of additional birth defects, geographical areas and other factors[20]. Potential explanations given for the predominance among females include differences between the sexes in embryonic developments' susceptibility to teratogenic insult and spontaneous abortion rates[21]. However, amidst such evidence, it is interesting to note that our study cohort presents contrasting results of male predominance in the management. With such distinct evidence, it is captivating to highlight that some sub-Saharan African countries like Nigeria and Cameroon have presented results showing male predominance during the management,[21,22] hence our results are in line with such findings. Probably an explanation can be connected to ethno-theories where boys are much more treasured than girls in most societies in sub-Saharan Africa. For instance, in the Zambian society, it has been observed that women with disabilities are described as 'role-less' and regarded as unfit in terms of being sexual partners thus women with physical disabilities are viewed as having a 'double handicap'[23].

The results of the current study are in line with the pattern of reports given by some countries of sub-Saharan Africa where 56% of the children went for surgical repair after 48hours of life. The incidence of live births with SB in the developed countries has diminished in the recent years through a combination of folic acid dietary supplements and abortions, thus neonates with SB are likely to be operated within 24-48 hours to repair the defect[24]. On the other hand, the story of the sub-Saharan African countries is that of prevailing low socioeconomic status, harmful taboos, religious beliefs, paucity of medical manpower and inadequate facilities which make late presentation of patients the norm, with children typically presenting after the first week of life[25].

It was eminently seen that 61% of patients were diagnosed with hydrocephalus as an associated problem while 39% had SB alone. Studies have shown that hydrocephalus occurs in association with SB[6-8,17,26] in about 80-90% but less likely to occur in those with lower spinal lesions[4]. Our results are in support of the notion that hydrocephalus occurs in association with SB because 61% of the children with SB in the study sample presented with hydrocephalus.

Currently, available data in the Ministry of Health is limited to support the assertion that folate deficiency is common in Zambia[27]. The results of the study showed that children with SB are being born in Zambia, although there is a great possibility that a great number of children are still locked up in the homes and others die in their homes without the Ministry of Health knowing. In spite of known scientific basis of the need for folates in the closure of the neural tube and the evidence of children born with NTDs everyday in Zambia, there is no program targeted at improving the nutrition of young women early in life in order to consolidate healthy eating habits and life style behaviours[27]. Consequently, inadequate attention is accorded to issues of maternal nutrition, infant and child survival, thus compromising women's nutritional status, thus not contributing to the global turn down of NTDs. In order to minimize the levels of NTDs, and maternal and neonatal death, a variety of preventive measures and life-saving interventions are needed at all stages of pregnancy, delivery, and post-partum and at many different levels. It is quite unfortunate that NTDs which are irreversible occur even before the expectant mother even becomes aware of the developing pregnancy[28].

It has been reported globally that myelomeningocele is the most common defect in SB[21,26]. This postulation has been supported by a prospective study done in a large cosmopolitan Western African city which showed that the most common defect in the children with NTDs was myelomeningocele[19] although in contrast, another study was done on spina bifda cystic in Zaria, Nigeria that showed more children with meningocele compared with myelomenigocele[25]. Our findings showed that myelomeningocele was the most common diagnosis in the treatment cohort hence supporting the reports attesting that myelomingocele is the commonest defect.

There seem to be contrasting views on the common site of the defect in SB. For instance Campbell and others[26] report that approximately 80% of all lesions in infants with SB occur in the lumbar region and others also have subscribed to the same concept and went on to justify that the lumbar region is the last region of the neural tube to close[29]. Conversely, some studies have found that the lumbo-sacral region is the commonest site for SB[25,30,31] and a study done in Zambia showed the sacral region to be the most common site of occurrence[32]. However, our results support the notion that the lumbar region is significantly, the common site being affected although dissimilar with other findings.

Our findings also showed that Southern province had more children that originated from there than any other province in the country. Southern province is one of the provinces with a high number of health facilities thus making probable speculations very difficult[27]. Some of the probable likely facts that need investigation could be the impact of early and late child birth, toxoplasmosis, use of cow gung, droplets from bats mixed with soil eaten by expectant mothers, dependency on maize and milk and particular wild fruits eaten there.

Interventions given to the children

In Western medical centers, emphasis has been placed on simultaneous myelomeningocele closure and ventriculoperitoneal shunting for children with SB and co-morbid hydrocephalus. This is not practical in developing countries where patients present in a delayed fashion, many with open, dirty myelomeningoceles. Peculiar harsh economic and social realities make late presentation, malnutrition and sepsis at presentation prevalent. For instance, studies done in Nigeria, Cameroon, Kenya and Uganda on the management of SB have shown post-operative complications of leaking cerebrospinal fluid and wound infections[21,22,25,30]. Our results reported cerebrospinal fluid leak and superficial site infections in line with reported surgical complications that have been previously reported in some African countries.

Early third ventriculostomy and CPC are being recommended as the current surgical options for hydrocephalus management considering problems in developing countries because of the decrease of the number of hospital days and emergency visits for shunt failure[7,33]. The successful procedure of ETV-CPC has been reported in West Africa in Lagos, Nigeria[30] and East Africa in Mbale, Uganda where children treated came from Kenya, Tanzania, Malawi, Somalia, Rwanda, Congo and Mauritius[2].

Several studies have shown that physiotherapy interventions help to improve mobility and functional independence in children with SB[34-38]. It is quite sad to note that physiotherapy services were not very much utilized. However, it has been observed that physiotherapists spend more time with children with SB. Furthermore, it has been stated that the concept of sensitive periods assumes that children are more responsive to experiential learning during the first three years of life when there is rapid brain growth and plasticity[39] hence utilization of physiotherapy can help the children quite a lot in the follow up program.

Surgical and ongoing physiotherapy treatment of SB and associated hydrocephalus has ensured long-term survival in patients selected for treatment. Continued care of these patients, however, is a long-term process with the attendant problems of motor deficits, incontinence, cognitive impairment and psychosocial adjustment[40]. The results of our study showed that only a few of the children were compliant to the treatment program. Additionally, from the treatment period, we noted that most children were only managed in the specialized hospitals for the first year of their life and later may have probably made use of their local hospitals. An explanation to our current results could be attributed to the findings of Munkonge and others[41] who conducted a study to determine the compliance with the reviews after insertion of the shunts and discovered that southern province had a higher prevalence compared with other provinces. The majority of mothers revealed that they did not take their children for review because of financial problems.

It was observed that a good number of children were subjected to HIV testing hence parents were counselled before the test was done in preparation for surgery. There is scarcity of research that has been done on the interplay between HIV/AIDS and disability in Zambia or internationally. The complex dynamic between HIV/AIDS, disability and poverty in sub-Saharan Africa requires a more thorough look in order to really understand how these may self-replicate and reinforce one another. The link between poverty and disability has been widely documented and is widely recognized that povery causes disability and disability causes poverty particularly in resource limited settings[42]. However research results on the relationship between disability and HIV/AIDS shows that the AIDS epidemic has been largely unrecognized among both disability and advocacy groups[43]. Knowledge of the HIV status helps in the management of children in order to be cautious about certain eventualities that may arise due to the impact of the virus on the outcomes of surgery and consequently on the healing mechanisms.

Instruments used for measuring outcomes of interventions

A good number of studies have assessed long-term outcomes of individuals with SB with respect to physical function, cognitive ability, psychosocial

development and the impact on the family[9,44-53]. These studies suggest among other things that SB has an impact on the functional activities of the individual in particular self-care, mobility and social function. Our results showed that self-care, mobility, social function, continence and stretching and POP casting for foot correction were considered in the management. This is supported by studies that have highlighted the importance of the three main domains of care being self-care, mobility and social function.

Although a good number of studies have been done on outcomes, there are only two paediatric tools used to assess functional outcomes: the Paediatric Evaluation Disability Inventory (PEDI) and the Functional Independence Measure for Children, (WeeFIM). Additionally, both instruments were never constructed for children with SB, but for children with disabilities in general. Furthermore, both the instruments were developed in America and Europe and there is no evidence of an instrument that was constructed in Africa for children with SB[9,26,54-57]. Therefore, it was not so surprising that none of the 253 files of children had any kind of instrument measure that was used to evaluate the impact of interventions given, except for the standardized assessment forms for serial head circumference measurements in those children that had hydrocephalus as well. This practically shows that effects of interventions given to children with SB were not measured.

STUDY LIMITATION

Tracing files at the UTH was a great challenge. There is a possibility that some files may not have been identified. The interchange of patients between the two hospitals was also a challenge because of lack of a proper system of tracking files. A high number of children were lost to follow-up hence making it difficult to know the proper outcomes of the interventions given to the children.

CONCLUSIONS

Rehabilitation of children with SB in Zambia posses some great challenges due to the so many logistical problems connected with limited specialized facilities only found in Lusaka. There is male predominance in the children being managed with SB and the children present late for surgical interventions. Evidence has been provided that hydrocephalus occurs in association with SB. Myelomeningocele is the most common defect among children with SB and the most common site of defect is the lumbar region of the spinal cord. It has been eminently observed that most of the children with SB came from the Southern Province of Zambia.

Physiotherapy was under-utilized and generally, there was non-compliance in almost all the children. It is difficult to tell the impact of interventions given to children because there are no measuring instruments available. It has been noted that clinical knowledge and scientific understanding among care providers in Zambia is there, but without proper follow-ups, it becomes difficult to justify outcomes especially if they are not even properly defined. As Anderson[58] puts it in his analogy that, '*Whilst perhaps a truism, we need to remember that lack of evidence is not evidence of lack of effect'*, therefore problems of providing evidence may lead to the inability of appreciating the great efforts being made.

RECOMMENDATIONS

It is highly recommended that proper follow-up programs are instituted by both the UTH and BCH in the management of children with SB in Zambia. Physiotherapy services can be utilized for they can help provide a good network for follow-ups. It is highly recommended that a longitudinal study be done to investigate the predominance of the prevalence of SB in the Southern Province of Zambia. Researchers are challenged to develop measuring instruments that are culturally sensitive and appropriate to the needs of Zambian children.

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