Spina bifida in a Tertiary Health Institution: A 17-Year Experience

Sunday Patrick Nkwerem[1,2], Jude-Kennedy Chinedu Emejulu[1,2], Ofodile Ekweogwu[1,2], Tochukwu Henry Mbanugo[2], Onyemaechi Ereke Nwibo[2] and Ikechukwu Valentine Okpalike[2].

[1]Nnamdi Azikiwe University, Nnewi Campus. [2]Nnamdi Azikiwe University Teaching Hospital, Nnewi.

ABSTRACT.

Background: Split spine has shown a progressively changing temporal trend. It appears to be decreasing in frequency in advanced countries unlike what is observed in some Africa countries. It also appears to have different epidemiological patterns, based on ethnicity, race and geographic location. **Objectives:** This study aims to review the clinical profile of spina bifida patients that were operated on in a tertiary health institution in Anambra State between 2006 and 2023. **Materials and Methods:** It is a retrospective study. Records were obtained from patients' folders, theatre and ward admission registers. Demographic data like age and sex were collected, diagnosis, presenting symptoms, operation done and complications were retrieved using the data extraction template. The data was analysed with SPSS 23, while results were presented in tables and chats. **Results:** A total of 83 cases were operated on within the period. Average cases per year was 4.9 while almost half of the total number of cases were seen in the first four years of study. About 74% of the patients presented within the first two years of age. They were mostly males, while the most common type was spina bifida cystica. The most common region affected was lumbosacral. Eight presented with rupture. Twenty-eight patients had CSF diversion for hydrocephalus. Ten patients had surgical site infection, while thirteen mortalities were recorded. **Conclusion:** There is a decreasing frequency of patients with spina bifida in the study. Lumbosacral region is still the most affected region while spina bifida cystica is the most common variant.

Keywords: Bifida; Experience; Health; Institution; Spina bifida.

OPEN ACCESS

***Correspondence**:
*Dr Sunday Patrick Nkwerem.
Nnamdi Azikiwe University,
Nnewi Campus*
Email:
nkwerempatrick@gmail.com
***Tel**: +2348032927735*

***Specialty Section**:
This article was submitted to Clinical, a section of TJMR*

Article Metrics:
Submitted: 11 Octob 2024
Accepted: 29 Dec 2024
Published: Jan-June, 2025

***Citation**:
SP Nkwerem, J-KC Emejulu, O Ekweogwu, TH Mbanugo, OE Nwibo and IV Okpalike. Spina bifida in a Tertiary Health Institution: A 17-Year Experience. Trop J Med Res. 2025:24(1);37-43.
10.5281/zenodo.14659356.*

***Journal Metrics**:
ISSN: p1119-0388, e2505-0338*
***Website**: www.tjmr.org.ng*
***E-mail**: info.tjmr@gmail.com*

***Publisher**: cPrint Publishers*

Access Code

www.tjmr.org.ng

INTRODUCTION

Spina bifida literarily means split spine in Latin. It is a congenital neural defect in which there is incomplete closure of the spine and the membranes surrounding the spinal cord during the first month of pregnancy. It is a major cause of preventable disabilities in children. [1,2] It is also a major cause of psychological distress to both patients and caregivers alike.[3]

Diverse epidemiological patterns have been noted based on ethnicity, race, geography and temporal trends.[4] The prevalence of spina bifida in certain 10 regions of the US was put at 3.1 per 10000.[5] The pool prevalence rate of spina bifida in Africa is 0.13% with a range between 0.12% and 0.14%.[6] In Kenya, Githuku *et al* recorded 4.4 per 10,000 live-births for both spina bifida and encephalocele.[7] Uba *et al* found an incidence of 0.5/1000 live births and 1.9 of all admissions in North Central part of Nigeria.[7] There appears to be some variation in the gender distribution in different regions.[7-9] The interplay of genetic and environmental factors is noted in the aetiopathogenesis of spina bifida. Spinal bifida has been noted to be more common in trisomy 13, and trisomy 18.[4] An increased association has also been noted in Acro-callosal syndrome, CHILD syndrome, Fraser syndrome, Waardenburg syndrome and Meckel-Gruber syndrome, as well as family history.[4] Relative lack of function of folic acid is perhaps one of the well documented environmental aetiological factors.[10] It is however believed to be due to mutation in the genes for folate-homocysteine pathway especially methyl tetrahydrofolate reductase (MTHFR). Administration of therapeutic high doses of folic acid is presumed to be able to overcome the relative defect in function of this enzyme. Maternal diabetics mellitus has also been associated with spina bifida.[11] Other risk factors identified include intrauterine exposure to carbamazepine, valproic acid and nonsteroidal anti-inflammatory drugs (NSAIDs). [12,13]

Beyond clinical evidence of neurological weakness and loss or poor sphinteric control, spina bifida may occasionally be associated with hydrocephalus.[14] Excision and repair are surgical procedure often needed for infection prevention, cosmesis and untethering of the cord.

From the foregoing, it is evident that distribution of spina bifida varies with ethnicity, race, geographic distribution and temporal trends.[4,6,15-17] Furthermore the study of congenital anomalies in Enugu did not elaborate the clinical profiles, while there is no data on the above anomaly in Anambra in the last ten years.[18-20]

Objectives: This study aims to review the clinical profile of spina bifida patients that were operated on in a tertiary health institution in Anambra State between 2006 and 2023.

PATIENTS AND METHODS

Study design/area/population

This is a retrospective study of all the cases of spina bifida patients operated by the three neurosurgical units of Nnamdi Azikiwe University Teaching Hospital, Nnewi between 2006 and 2023. The three neurosurgical units currently handle spina bifida.

Inclusion criteria

All patients operated on by the three neurosurgical units.

Exclusion criteria

Those who were attended to by the units but could not be operated on.

Sampling approach

Convenience sampling method was deployed

Procedure: Data were retrieved from both patients' folders; theatre register and ward admission register using data extraction template. Demographic data like age and sex were collected, diagnosis, presenting symptoms, operation done and complications were also retrieved.

Statistical analysis

The data was analysed with SPSS 23 [IBM Corp, Armonk NY], while results were presented in tables and chats.

Ethical consideration

Ethical clearance was obtained for the study.

RESULT

A total of 83 cases were operated on within the period under review. Average cases per year was 4.9 while almost half of the total number of cases were seen in the first four years of study. About 74% of the patients presented within the first two years of age (see table 1).

Table 1: Age Distribution
N = 83

Age (months)	Frequency	Percentage (%)
0-12	41	49
>12-24	19	23
>24-36	9	11
>36-48	8	10
>48-60	2	2
>60	1	1

Most patients presented within the first year of life. There was male preponderance (59%). The most common type was spina bifida cystica (see figure 1)

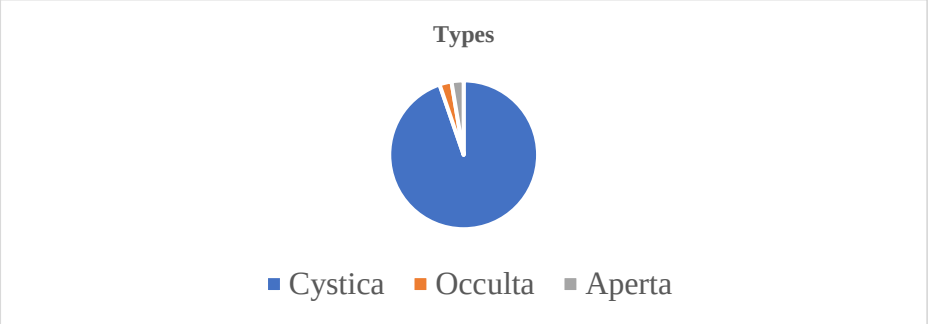

Figure 1: Spina cystica was the most common type

The most common region affected in the study was lumbosacral region (see figure 2)

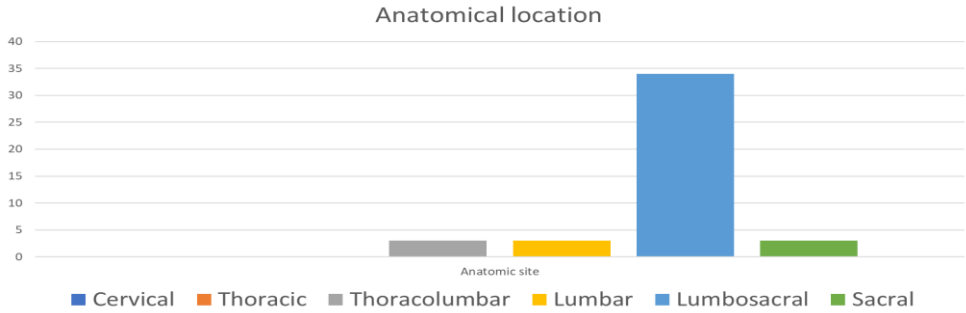

Figure 2: Lumbosacral region was the most common anatomical distribution

Eight cases were ruptured at presentation. Twenty-eight cases needed CSF diversion. All cases had excision and repair. Ten patients had surgical site infection while seven had post operative sepsis. Thirteen mortalities were recorded.

DISCUSSION

Temporal and regional epidemiological patterns have been noted in distribution of spina bifida.[4-7] This may be attributed to the improving diagnostic tools, surgical services and increasing manpower in some regions. In the index study, 83 cases were operated on in the said period by the neurosurgical team. These however, excludes cases that did not

end up in the theatre for various reasons. In the same centre, 44 cases were attended to (both those operated on and those that could not be operated on) over 44 months.[20] In Enugu, Ozor *et al* found 47 patients in two tertiary institutions in a five-year observational study.[18] A single institutional retrospective study in the same city, Enugu found 49 cases over 42 months of study. This trend appears to be different in North West where 90 cases were managed in one year and South West Nigeria where one hundred and six cases were managed over fourteen years.[16,21] In Jos, North Central Nigeria, 284 cases were attended to in 17 years.[8] Shehu *et al* managed 77 cases in 11 years in Zaria, Nigeria.[22] Different patterns have been noted across Africa. [6] The difference in methodology may partly account for these differences.

There appears to be more cases of spina bifida operated on in the first four years of institutional practice and a temporal trend of decrease in the recent years. This may be accounted for by the improving antenatal care, increasing awareness of the benefit of periconceptional folic acid. More centres (private and public institutions) in the state now attend to spina bifida.

An increasing pattern of presentation was noted in Africa between 1983 to 2018.[23] This is against the decreasing trend observed in developed countries.[24-26] This decreasing temporal trend has been attributed to the increasing understanding of the aetiology and increased used of periconceptional folic acid.[20,24,26,27] The poor indices of spina bifida in Africa may be connected with the poverty, level of education, poor health facility, HIV infection and poor implementation of periconceptional folic acid.[23]

Most of the patients that were operated on in this study presented within the first year of life while cumulatively, about 72% presented within the first two years of life. In Enugu, Eke *et al* found most patient with spina bifida to be below 1 year at presentation.[19] Though the sample population included people with other CNS anomalies. In Zaria, most of the patients were less than six months of age.[22] The associated social consequence on the parents may be responsible for the early presentation of spina bifida. The increasing availability of neurosurgical services and public awareness may have contributed to early referral and early presentation to the appropriate centres for care.

This study showed male preponderance which is similar to studies by Ozor *et al* and Eke *et al* in Enugu.[18,19] In Ife, South West Nigeria, Alatise *et al* found male preponderance.[21] Though a more recent study, by Komolafe *et al*, in the same centre was almost of equal gender distribution.[24] This pattern may be explained by the fact that congenital anomalies especially congenital CNS anomalies are more common in males.[19,25,26] Spina bifida cystica is the most common type seen in the study. It accounted for over 94 % of cases in this study. This is similar to an earlier study in the centre and also in other parts of the world.[6,18] The most common anatomic location, in this study was the lumbosacral spine (80%). Similar pattern was observed by other workers across Nigeria. [8,16,21,22,28] Eight patients (10%) presented as an emergency with the rupture of the cyst. Uba *et al* found a higher incidence of ruptured spina bifida (42%) in North central Nigeria.[8] In Uganda, Byabato *et al* found the incidence of ruptured myelomeningocele to be slightly over 25%. The higher rate of rupture at presentation, in the Northern Nigeria, may not be unconnected with high poverty rate, low literacy as well as poor health seeking behaviour.

About 34 % of the patients in this study had CSF diversion. Alatise *et al* found a higher incidence of hydrocephalus (53.8%) in their study. Uba *et al* even had higher incidence of hydrocephalus. [8] In North West Nigeria, Obanife *et al* noted that about 45.6% of patients had hydrocephalus.[16] These geographic differences may be connected with the methodology. For example, Obanife *et al* made the diagnosis of associated hydrocephalus in spina bifida patients with trans fontanelle ultrasound while in the index study, the number of patients with hydrocephalus was based on the number of patients documented to have had CSF diversion.

All the patients had the standard treatment which was excision and repair of the spina bifida. About 13% of cases had local surgical site infection postoperatively while 11% had sepsis. This is comparable to the findings by *Uba et al*. Shehu *et al* found similar pattern of surgical site infection in Zaria.[22] The mortality rate in this study was 17%. Alatise *et al* found a higher mortality rate of 27%. In countries like USA and South Africa, the mortality

was markedly lower.[29,30] This may be attributed to better economies, higher literacies and better health facilities.

Being a retrospective study with a relatively small sample size is a limitation to the study as this makes strong inference from the study difficult, and these are major limitations to the study. In addition, some patients did not have proper documentation of all the parameters investigated. Furthermore, the study sample was only patients operated on in the above centre thereby introducing some element of bias as patient who presented to this institution but had to be operated in another centre because of various reasons were not included in this study. A well-coordinated multicentre study will give a better picture of the current state of spina bifida in the region and in the country. Despite these, this study has been able to show some pattern of distribution and clinical profile of spina bifida operated on in the last seventeen years of the centre of study.

CONCLUSION

There is decreasing number of patients with spina bifida attending the studied facility. Most of the patients present in the first year of life. Spina bifida cystica is the most common type and lumbosacral region is the most frequent anatomic location.

Acknowledgement

We deeply appreciate the resident doctors and nursing staff who were part of the care of these patients.

Authors contribution

SPUN conceptualized and designed the study. JKCE, OEN and IVO contributed to implementation of the project and revision of the manuscript. All authors were involved in the writing and revision of the manuscript. The authors read, approved the final manuscript and agree to be accountable for all aspects of the work.

Data availability

The data used to support the findings of this study are available from the corresponding author upon reasonable request.

Funding

No funding sources.

Conflict of interest

None declared.

Ethical approval

The study was approved by the Institutional Ethics Committee.

REFERENCES

1. Ledet III LF, Plaisance CJ, Daniel CP, Wagner MJ, Alvarez I, Burroughs CR, et al. Spina Bifida Prevention: A Narrative Review of Folic Acid Supplements for Childbearing Age Women. Cureus. 2024;16(1).
2. Grosse SD, Berry RJ, Tilford JM, Kucik JE, Waitzman NJ. Retrospective assessment of cost savings from prevention: folic acid fortification and spina bifida in the US. American Journal of Preventive Medicine. 2016;50(5):S74-80.
3. Lidal IB, Lundberg Larsen K. Anxiety, depression, and fatigue in middle-aged and older persons with spina bifida: a cross-sectional study. Disability and Rehabilitation. 2022;44(25):7936-46.
4. Williams LJ, Mai CT, Edmonds LD, Shaw GM, Kirby RS, Hobbs CA, et al. Prevalence of spina bifida and anencephaly during the transition to mandatory folic acid fortification in the United States. Teratology. 2002;66(1):33-9.
5. Shin M, Besser LM, Siffel C, Kucik JE, Shaw GM, Lu C, et al. Congenital Anomaly Multistate Prevalence and Survival Collaborative. Prevalence of spina bifida among children and adolescents in 10 regions in the United States. Pediatrics. 2010;126(2):274-9.
6. Oumer M, Taye M, Aragie H, Tazebew A. Prevalence of Spina Bifida among Newborns in Africa: A Systematic Review and Meta-Analysis. Scientifica. 2020;2020(1):4273510.
7. Githuku JN, Azofeifa A, Valencia D, Ao T, Hamner H, Amwayi S, et al. Assessing the prevalence of spina bifida and encephalocele in a Kenyan hospital from 2005–2010: implications for a neural tube defects

surveillance system. The Pan African Medical Journal. 2014;18.
8. Uba AF, Isamade ES, Chirdan LB, Edino ST, Ogbe ME, Igun GO. Epidemiology of neural tube defects in North Central Nigeria. African Journal of Paediatric Surgery 2004; 1 (1): 16-19
9. Alageb NA, Satte M. Epidemiology and risk factors of spina bifida in Sudan. Journal of Global Biosciences. 2020;9(6):7528-35.
10. Oakley Jr GP. The scientific basis for eliminating folic acid–preventable spina bifida: a modern miracle from epidemiology. Annals of epidemiology. 2009;19(4):226-30.
11. Mitchell LE, Adzick NS, Melchionne J, Pasquariello PS, Sutton LN, Whitehead AS. Spina Bifida. Lancet 2004;364:1885–95
12. Esposito DB, Parker SE, Mitchell AA, Tinker SC, Werler MM. Periconceptional nonsteroidal anti-inflammatory drug use, folic acid intake, and the risk of Spina Bifida. Birth defects research. 2021;113(17):1257-66.
13. Koren G, Nava-Ocampo AA, Moretti ME, Sussman R, Nulman I. Major malformations with valproic acid. Canadian family physician. 2006;52(4):441-2.
14. Blount JP, Maleknia P, Hopson BD, Rocque BG, Oakes WJ. Hydrocephalus in Spina Bifida. *Neurol India*. 2021;69(Supplement):S367-S371. doi:10.4103/0028-3886.332247
15. Nnadi DC, Singh S. The prevalence of neural tube defects in North-West Nigeria. Saudi Journal for Health Sciences. 2016;5(1):6-10.
16. Obanife HO, Nasiru IJ, Lagbo J, Otorkpa EJ, Shehu BB. Epidemiology of myelomeningocoele in Africa: An experience in a Regional Neurosurgical Center in Northern Nigeria. Journal of Pediatric Neurosciences. 2021:10-4103.
17. Ismail NJ, Lasseini A, Koko AM, Shehu BB. Clinical Profile of Spina Bifida Cystica: Six-Year Review from a Regional Centre, in Nigeria. Age (months).;1(5):6-12.
18. Ozor II, Chukwubuike KE, Enyi N. Profile of Neurological Congenital Anomalies in the Two Teaching Hospitals in Enugu, Nigeria. J. Neuroscience and Neurological Surgery. 2021:9(1); DOI:10.31579/2578-8868/175
19. Eke CB, Uche EO, Chinawa JM, Obi JE, Obu HA, Ibekwe RC. Epidemiology of congenital anomalies of the central nervous system in children in Enugu. Nigeria: A retrospective study. Ann Afr Med;2016 15:126-32.
20. Emejulu JK, Okwaraoha BO. Peculiarities in cases of spina bifida cystica managed recently in south-east Nigeria: could antimalarial drugs be a major but unrecognized etiologic factor?. *Pediatr Neurosurg*. 2011;47(3):194-197. doi:10.1159/000334255
21. Alatise OI, Adeolu AA, Komolafe EO, Adejuyigbe O, Sowande OA. Pattern and factors affecting management outcome of spina bifida cystica in Ile-Ife, Nigeria. *Pediatr Neurosurg*. 2006;42(5):277-283. doi:10.1159/000094062
22. Shehu BB, Ameh EA, Ismail NJ. Spina bifida cystica: selective management in Zaria, Nigeria. *Ann Trop Paediatr*. 2000;20(3):239-242. doi:10.1080/02724936.2000.11748142
23. Ssentongo P, Heilbrunn ES, Ssentongo AE, Ssenyonga LVN, Lekoubou A. Birth prevalence of neural tube defects in eastern Africa: a systematic review and meta-analysis. *BMC Neurol* 22, 2022: 202. https://doi.org/10.1186/s12883-022-02697-z
24. Ba G, Wu QJ, Chen YL, Huang YH, Gong TT. Prevalence and time trends of spina bifida in fourteen cities located in the Liaoning province of northeast China, 2006-2015. *Oncotarget*. 2017;8(12):18943-18948. doi:10.18632/oncotarget.14848
25. Liu J, Zhang L, Li Z, Jin L, Zhang Y, Ye R, Liu J, Ren A. Prevalence and trend of neural tube defects in five counties in Shanxi province of Northern China, 2000 to 2014. *Birth Defects Res A Clin Mol Teratol.* 2016;106:267–274.
26. Atta CA, Fiest KM, Frolkis AD, Jette N, Pringsheim T, St GC, et al. Global Birth Prevalence of Spina Bifida by Folic Acid Fortification Status: A Systematic Review and Meta-Analysis. *Am J Public Health.* 2016;106:e24–e34.
27. Rodrigues VB, Silva END, dos Santos AM, Santos LMP. Prevented cases of neural tube defects and cost savings after folic acid fortification of flour in Brazil. PLoS ONE

2023; 18(2): e0281077. https://doi.org /10.1371/journal.pone.0281077

28. Komolafe EO, Onyia CU, Ogunbameru IO, Dada OA, Owagbemi OF, Ige-Orhionkpaibima *et al*. The pattern, peculiarities, and management challenges of spina bifida in a teaching hospital in Southwest Nigeria. *Childs Nerv Syst*. 2018;34(2):311-319. doi:10.1007/s00381-017-3614-8
29. Mashiloane, P C, & Masekela, R. A review of the epidemiology, post-neurosurgical closure complications and outcomes of neonates with open spina bifida. South African Journal of Child Health. 2020;*14*(2)77-81. https://dx. doi.org/10.7196/SAJCH.2019.v14i2.1638.
30. Dicianno, BE. Sherman A, Roehmer C, Zigler CK. Co-morbidities Associated With Early Mortality in Adults With Spina Bifida. American Journal of Physical Medicine & Rehabilitation 2018; 97(12):861-865. | DOI: 10.1097/PHM.0000000000000964.