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Original Research Article

Demography and management outcome of neural tube defects in a Nigerian tertiary health institution

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ABSTRACT

Background: Neural tube defects (NTD) are a group of congenital anomalies of the central nervous system (CNS). Its management is very challenging. A recognised leading cause is a folic acid deficiency, therefore prevented by taking a preconception folic acid. This study looked at the demographic features and management outcome of neural tube defect, a fairly common preventable condition with the need to raise awareness on its preventive measures.
Methods: The study was conducted on 82 patients with NTD that were managed in federal medical centre Yola, in North-Eastern Nigeria. It was a retrospective study over a 4-year period, from January 2016 to December 2019.
Results: Ages at presentations ranged from 1 to 93 days with a mode of 3 days. There were 29 males and 53 females with a male to female ratio of 1:1.8. Spina bifida constituted 74.4%, encephalocele (20%), Anencephaly (5%). Types of spina bifida managed were myelomeningocele (63.5%), meningocele (33.3%), and lipo-myelomeningocele (3.2%). Two had a 2-level meningocele. Syndromic associations of myelomeningocele were with hydrocephalus (78.1%), foot deformity (22.2%), cardiac (2.5%), and anorectal malformation (2.5%). Ninety-five-point one had various surgeries with 30.8% developing postoperative complications. commonest was post excision of myelomeningocele and encephalocele hydrocephalus (29.1%) in 7 patients. Post-operative Mortality was reported in 2 patients (8.3%).
Conclusions: Myelomeningocele was the commonest NTD, and its syndromic association with hydrocephalus was common. A large number of patients had surgical interventions with a good outcome. Post-op mortality was minimal.

Keywords: Demography, Management, Outcome, NTD

INTRODUCTION

Neural tube defects (NTD) are developmental defects of the CNS seen at birth. The CNS develops from the neuroectoderm, in the third week of gestation, following its folding and subsequent formation of a tube-like structure (Neural tube).[1] The anterior (rostral) and posterior (caudal) end usually closes respectively by 21 and 28 days after conception, with the closure beginning in the middle and extending in cranial and caudal directions.[1] From the rostral part, the brain (forebrain, midbrain, and hindbrain) develops, while the caudal part forms the spinal cord.[2] NTD may involve the whole length in a condition called posterior rachischisis, may remain open in the region of the brain because of the non-closure of the anterior neuropore, resulting in a condition called anencephaly or the region of the spine (Spina bifida).[3] Consequently, this may also result in the neural tissue lying outside the cranial cavity, or the vertebral canal. When this happens in the region of the brain the condition is called encephalocele or meningoencephalocele, and when it occurs in the spinal cord region it is called myelocoele or meningomyelocele (also called myelomeningocele). However, When the defect contains meninges without any neural tissue, it's referred to as meningocele irrespective of its location.[3]

The implicated causative factors of NTD include genetic factors, nutritional factors, environmental factors, and second-hand smoking or a combination of these, which are known to play a role in the development of NTD.[4,5] Global estimate is put at about 300,000 babies born with NTDs annually.[6] Reported incidences are about 0.6/1000 births in Africa but as high as 10/1000 birth among Arab countries.[7,8] The prevalence of NTD in Nigeria ranges between 0.95 and 7 per 1,000 births.[9,10]

It is a known fact that folic acid deficiency, a nutritional factor is said to play a leading role in the aetiology of NTD, and may be prevented by using preconception folic acid based on many studies.[11,12]

NTD may result in a variable degree of paralysis, developmental delays, lifelong disability and death with other associated complications that include hydrocephalus in patients with spinal bifida or encephalocele and congenital talipes equinovarus (CTEV).[13,14]

Socially, these patients often lack acceptance with severe stigmatisation in their communities with resultant social, economic and emotional distress to their families. Only 21% of them can walk independently. However, about half of those with spinal bifida and encephalocele (51%) cannot walk.[15]

The risks of having a child with NTDs increases from 2 to 3% if one pregnancy has been affected, and to 10% if two pregnancies have been affected.[16]

To date, no similar study has been carried out in our facility. Therefore, we aimed at identifying the important demographic factors, the spectrum of NTD, the available interventions, and the complications that ensued. Our findings may bring forward NTD as a public health problem, the availability of interventions in our region and the need to adopt proactive preventive measures.

METHODS

We carried out a retrospective study on all children who presented with NTD to federal medical centre, a tertiary medical centre located in Yola, Adamawa State, Nigeria, over a 4-year period, from January 2016 to December 2019. We collected the patient's demographic data, treatment and complications that were observed. These include age at presentation, sex, type of lesion, level of the lesion, other associated anomalies, treatment offered, outcome, and complications that followed the surgical intervention. Only patients who had surgery with/without other intervention were considered for the study. Ninety-one patients with NTD were identified. Nine patients were excluded from the study because they were either lost to follow up or declined any form of intervention. Data were analysed using statistical package for social sciences (SPSS) version 20.0 (Chicago, IL, USA) for windows. Analysis was carried out for descriptive statistics and illustrated as proportions and percentages.

RESULTS

A total of 82 patients were included and evaluated during the study period. Ages at presentation ranged from less than 1 day to 93 days, with a mode of 3 days. There were 15 encephalocele (17.8%), 63 spina bifida (75%) with 2 patients having 2- level lesions and 4 Anencephaly. Three encephalocele and 36 spina bifida presented within 2 days of delivery. Three anencephalies presented on the 1st day of delivery (75%) while the other 1 presented on the 3rd day. Of 36 spina bifida patients that presented within 2 days of delivery, 30 were cases of myelomeningocele. However, only 5 of myelomeningocele patients presented within 8 hours of delivery, and this included 3 with ruptured lesions following spontaneous vaginal delivery, the other 2 were not ruptured (Table 1).

There was a preponderance of females, revealing 29 (35.4%) males and 53 (64.6%) females, with a male: female ratio of 1:1.8. The females outnumbered the males in all the clinical subtypes of anencephaly, encephalocele and spina bifida as shown in Table 1.

Sixty-three (75%) spina bifida lesions were found on the 61 patients. Sixty-one of lesions were overt, this included 40 myelomeningocele (63.5%), 21 meningocele (33.3%). While remaining 2 had occult (Lipo-myelomeningocele) lesions (3.2%). There were 17 cases of encephalocele and 4 anencephalies as shown in Figure 1.

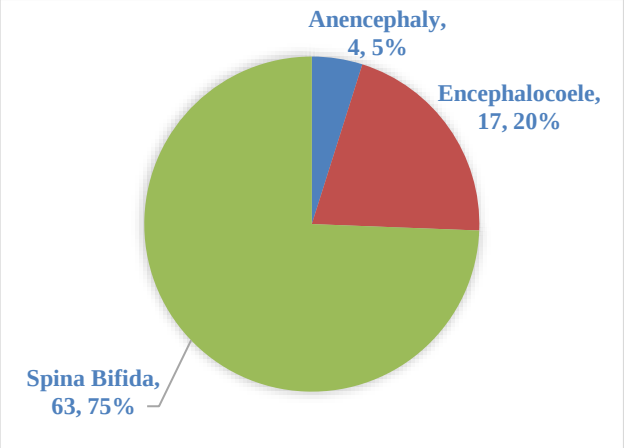

Figure 1: The distribution of the NTD lesions, (n=84).

Three (3, 17.6%) of the encephalocele were sincipital while 14 (82.4%) were occipital in location. Among the 14 occipital encephalocele, 3 were giants (21.4%) while the 11 were of lesser variable sizes.

Forty-three (68.2%) of the spina bifida were located in the lumbosacral region, consisted of myelomeningocele (72%), meningocele (23.3%), and lipo-myelomeningocele (4.7%) (Table 2).

Table 1: Ages at presentation and sex, (n=82).

Variables	Types of lesions (n=82), (%)			Number (%)
	Anencephaly	Encephalocele	Spina bifida	
Age at presentation (Days)				
≤2	3 (3.7)	4 (4.9)	36 (43.9)	43 (52.5)
3-29	1 (1.2)	6 (7.3)	15 (18.3)	22 (26.8)
≥30	0 (0)	7 (8.5)	10 (12.2)	17 (20.7)
Sex of the patients				
Male	1 (1.2)	7 (8.5)	21 (25.6)	29 (35.3)
Female	3 (3.7)	10 (12.2)	40 (48.8)	53 (64.7)

Table 2: Spina bifida types and levels, (n=63).

Types of spina bifida	Levels of the lesions				Total number (%)
	Cervical	thoracic	Lumbar	Lumbosacral	
Overt					
Meningocele	3	3	5	10	21 (33.3)
Myelomeningocele	0	0	9	31	40 (63.5)
Occult					
Lipo-myelomeningocele	0	0	0	2	2 (3.2)
Total number (%)	3 (4.8)	3 (4.8)	14 (22.2)	43 (68.2)	63 (100)

Two patients had 2 levels of meningocele in the following combination of cervical with lumbosacral and a thoracic with lumbosacral respectively.

Syndromic myelomeningocele was found in 34 (55.7%) of the spina bifida patients. These were with hydrocephalus (27, 78.1%) and foot deformity (8, 22.2%). The ones with Hydrocephalus exhibited features of Arnold-Chiari malformation (Table 3).

Table 3: Syndromic associations of myelomeningocele.

Types of associations, (n=34)	Number (%)
Foot deformity only	5 (14.7)
Hydrocephalus only	24 (70.6)
Foot deformity and hydrocephalus	3 (7.5)
Cardiac anomaly	1 (2.5)
Anorectal malformation	1 (2.5)

Some of the clinical findings of giant occipital encephalocele, a lumbosacral myelomeningocele with an anorectal malformation, and an intra-operative finding in a lipo-myelomeningocele is as shown below in Figure 2.

Seventy-eight patients had surgical excision and repair with ventriculoperitoneal shunt (VP shunt) for those with pre and post excision hydrocephalus. No form of intervention was done for the patients with anencephaly, all (4) died. The excision and repair surgery in 3 patients with sincipital encephalocele involved craniotomy, with a combined craniofacial repair. VP shunt was either done with excision at single sitting (2 in 1) in 20 (74.1%) patients while remaining 7 (25.9%) had it in 2 separate sittings following development of post excision hydrocephalus. The 42% of patients with foot deformity were attended to and treated by orthopaedic surgeons.

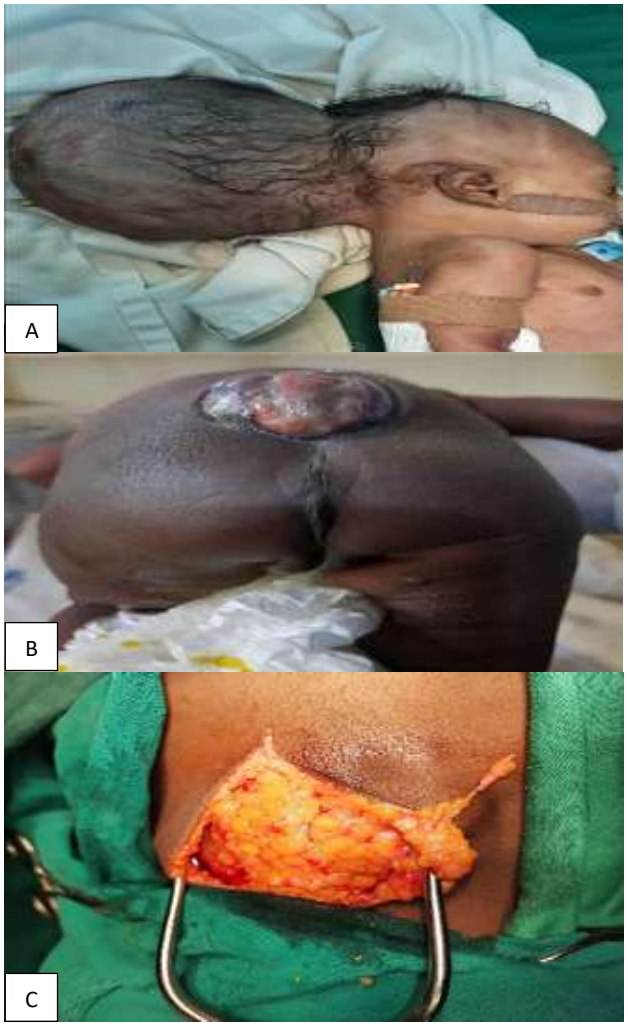

Figure 2 (A, B and C): A giant occipital encephalocele, lumbosacral myelomeningocele with imperforate anus, intra-operative lipo-myelomeningocele.

The post-operative complications were observed in 24 (30.8%) patients, these included post excision hydrocephalus, wound infections, CSF leak, hypertrophied scar, shunt obstruction, shunt infections, and post-operative mortalities (2, 8.3%) as shown below in Table 4.

Table 4: Post-operative complications and mortality, (n=24).

Types of surgery	Complications	Number (%)
Myelomeningocele excisions	Post excision HCP	5 (20.8)
	Wound infection	4 (16.7)
	Wound dehiscence	1 (4.2)
	Pseudo-meningocele	1 (4.2)
Meningocele excisions	CSF leak	3 (12.5)
	Hypertrophied scar	1 (4.2)
VP shunt	Shunt obstruction	3 (12.5)
	Shunt infection	2 (8.3)
Encephalocele excisions	Post excision HCP	2 (8.3)
	Mortalities	2 (8.3)

DISCUSSION

Half of our patients presented within 2 days of delivery. When compared to the findings from Lagos, Nigeria (50% within 2 weeks and Ife, Nigeria (52.8% in 1-7 days), ours presented earlier, dominated by the ulcerated lesion (Myelomeningocele) then, probably due to their ulcerated nature at birth.[17,18] None of the anencephaly patients was seen later than 3 days of delivery. This finding revealed an early referral of patients with an obvious life-threatening congenital anomaly in our setting, unlike other Nigerian findings of older patients in Lagos where 50% were ≤14-day olds and in Ife where 52.8% were less than 7-day olds respectively.[17,18]

We found a dominance of females over males in all the lesions. This is similar to the findings in Pakistan, Saudi Arabia, United States of America, South-Eastern Nigeria, Turkey and Suburban Nigeria.[18-24] However, others have reported male predominance in the studies from Uganda, Sudan, India, Kenya, Ife and Kano Nigeria.[18,25-27,29,30] Equal sex distribution was found in Jos and North Central Nigeria.[30,31]

Spina bifida was the commonest NTD, with its subsets of lumbosacral myelomeningocele being commoner than meningocele. Our findings are less than the reported 93% and 97% by Uba et al in North Central Nigeria and Githuku et al in Kenya respectively.[28,31] Conversely, much lower cases were reported by Ajah et al in South-Eastern Nigeria.[22] Among the overt spina bifida, most authors reported higher myelomeningocele than our findings as reported in Turkey (71%), Sokoto (84.6%) in Nigeria, Ife (86.8%) in Nigeria, and 92% in Uganda.[18,23,25,32] Despite our finding of relatively lesser meningocele, our finding is more than twice the findings of Ismail et al in Sokoto Nigeria and thrice the findings in Turkey and Ife in Nigeria.[18.23,32]

Based on spina bifida sites, lumbosacral lesions were more numerous than lumbar, thoracic and cervical lesions in descending order. Our finding on lumbosacral lesions is less than the reports from South-Eastern (80.6%) Nigeria, and in Sokoto (87%) Nigeria, but more than the finding from Sudan (53.3%) and North Central Nigerian by Uba et al.[22,26,31,32] The most common lumbosacral lesion was myelomeningocele, followed by meningocele with only 2 lipo-myelomeningocele. The finding of numerous lumbosacral myelomeningoceles conforms with many other reports.[20,26,31,32]

Anencephaly was not common, close to our finding was reported in Jos in Nigeria (3.7%), but lesser in Uganda that reported 1%.[25,30] Surprisingly Ajah et al in Southeast Nigeria reported an alarmingly high number (33.1%).[22]

We found that encephalocele constituted 20% of all the NTD, with occipital encephalocele more numerous than the sincipital type. Our finding is higher than the findings by Linda in Uganda (4%), by Githuku et al in Kenya (7%), Ajah et al (16.7%) in Nigeria and Turhan et al in Turkey (16%).[22,23,25,28] The finding of 25.9% in Jos Nigeria outnumbered ours.[30] Our number of giant encephalocele was slightly less than the finding of Bot et al in Sokoto (28%) Nigeria.[33] Although the study period and duration were not the same.

Syndromic associations are fairly common among patients with NTD (Spina bifida). The finding from Turkey by Turhan et al is very nearly similar to ours (56.5%).[25] Association of hydrocephalus with spina bifida in our finding was the commonest and is nearly consistent with findings in Sudan (65.7%) and North-Central Nigeria (68.3%).[26,31] However, findings in Ife (53.8%) Nigeria, South-Eastern Nigeria (14%) revealed lower associations, Kural et al and Laligan found higher associations of 83% and 90% respectively.[18,22,34,35] We found foot deformity to be fairly common among patients with myelomeningocele. A study in Lagos, Nigeria revealed the association of myelomeningocele with foot deformity to be the commonest.[17] Worthy of note that studies on foot deformities reported higher findings (30-50%) than ours as reported by Flynn et al, Westcott et al and Akbar et al.[36-38] However, a South-Eastern Nigeria study by Ajah et al revealed only 3% of foot deformities.[22]

We had a very high surgical intervention probably due to early referral and availability of neurosurgical care in most tertiary institutions in Nigeria, as compared to the interventions offered in Uganda, where 40% had excision and closure, and only 15% of the patients had VP shunt.[25]

Post-operative complications included hydrocephalus among others after excision of myelomeningocele and encephalocele, with the majority of hydrocephalus

following the excision of myelomeningocele. A Sudanese report gave 0.7% post excision hydrocephalus, which is very far less than our finding.[26] Overall mortality recorded was less than the report from Uganda that found 38%.[25] Both of our mortality was shunt related and recorded within 6 months of surgery, this is only about half of the findings of Khan et al and Reddy et al.[39,40]

This study was limited by being a retrospective one, characterised by few missing data in the patient's case files.

CONCLUSION

In conclusion, this study agreed with the fact that spina bifida is the commonest NTD, with myelomeningocele being commoner than meningocele. The syndromic association of myelomeningocele with hydrocephalus was also common. A large number of our patients presented fairly early and had surgical interventions with good outcomes. Post-operative mortality was minimal. From the study, it is worthy of note that NTD is of public health importance in our region and the need to put in place proactive preventive measures like folic acid supplements should be advocated.

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