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*Original Article*

# Surgical Management and Outcome of Neural Tube Defects in Children

*Das A[1], Alam MN[2], Rahman MS[3], Chowdhury MOA[4], Khan HU[5], Islam MS[6]

## Abstract

**Introduction: Neural tube defects (NTDs) are common congenital malformations that affect the entire central nervous system. Proper surgical management of affected children may lead to a meaningful and productive life, and deficiently managed cases of NTDs can be a catastrophic condition not only for patient but also for the patient's family.**

**Methodology: This prospective observational study was conducted in the Department of Paediatric Surgery, Sylhet M.A.G. Osmani Medical College & Hospital during the period from March, 2018 to August, 2020. Thirty children with a neural tube defect during the study period were included. Associated with life threatening major congenital malformations were excluded. The surgical management protocol used for different sites of NTDs was studied.**

**Results: The ages of the patients ranged from 1 day to 9 years with the median age of 3.5 days. There were 21 (70.0%) male and 9 (30.0%) female with a ratio of male to female was 2.33:1. Tendency to neural tube defect was more frequent in male than that of female (p=0.028). The types of NTDs were myelomeningocele (73.3%), encephalocele (10.0%), lipomeningocele (10.0%) and meningocele (6.7%). The sites of neural tube defects were lumbosacral (76.7%), occipital (10.0%), sacral (6.7%), lumber (3.3%) and cervical (3.3%). The nature of neural tube defects was ruptured in (60.0%) and not ruptured in (40.0%). Associated birth defects in this study were hydrocephalus in 56.7%, club foot in 16.7%, cervical prolapse in 6.7% and kyphosis in 6.7% cases. Associated neurological defects were lower limb paresis and paralysis in 83.3%, Urinary incontinence in 50.0% and faecal incontinence abnormality in 40.0% of cases. Postoperative complications were wound infection in 10.0%, wound dehiscence in 16.7%, CSF leakage in 23.3% and meningitis in 10.0% cases. The overall mortality rate was 6.7% of cases. Outcome of this study was recovery without complication in 66.7%, recovery with complications in 26.7% and death in6.7% of cases. Recovery without complication was more frequent than recovery with complications and death.**

**Conclusion: Lumbosacral myelomeningocele was the most common type of NTD. There was recovery without complication in most cases after operation. Early operation is associated with recovery without complication.**

**Keywords: surgical management of NTDs, outcome of NTDs, neural tube defects, myelomeningocele, encephalocele, meningocele**

1. Dr. Anupam Das, Assistant Professor, Department of Paediatric Surgery, Sylhet Women's Medical College Hospital, Sylhet, Bangladesh.
2. Prof. Dr. Md. Nurul Alam, Ex. Professor and Head, Department of Paediatric Surgery, Sylhet MAG Osmani Medical College Hospital, Sylhet.
3. Late Dr. Md. Shamsur Rahman, Associate Professor, Department of Paediatric Surgery, Sylhet MAG Osmani Medical College Hospital, Sylhet.
4. Prof. Dr. Md. Oyes Ahmed Chowdhury, Professor and Head, Department of Paediatric Surgery, Sylhet Women's Medical College Hospital, Sylhet.
5. Late Dr. Habib Ullah Khan, Assistant Professor, Department of Paediatric Surgery, Sylhet MAG Osmani Medical College Hospital, Sylhet.
6. Dr. Md. Sirajul Islam, Assistant Professor, Department of Paediatric Surgery, Sylhet MAG Osmani Medical College Hospital, Sylhet.

**Corresponding to:** Dr. Anupam Das, Assistant Professor, Department of Paediatric Surgery, Sylhet Women's Medical College Hospital, Sylhet, Bangladesh.

Special regards: Prof. Dr. Khandaker Abu Talha, Professor and Head, Department of Neurosurgery, Sylhet Women's Medical College Hospital, Sylhet, Bangladesh.

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## Introduction:

Neurulation, one of the earliest and most crucial events in human development, generates the neural tube, the rudiment of the entire adult central nervous system (the brain & spinal cord). NTDs account for the most common congenital anomalies of central nervous system and results from failure of the neural tube to close spontaneously between 3rd and 4th weeks of development.[1] The prevalence rate of NTDs in India is 7.48/1000 live births.[2] This is one of the serious congenital malformation responsible for infant disability, morbidity and mortality.[3]

NTDs may be classified as open or closed on the basis of the presence or absence of the exposedneural tissue. Open NTDs include anencephaly, spinal rachischisis or spina bifida aperta/cystica (i.e. myeloschisis, myelomeningocele and meningocele) and encephalocele. Closed NTDs include spina bifida occulta, lipomatous malformations (e.g. lipomas and lipomeningocele), split cord malformations, neurenteric cysts, dermal sinuses, tethered spinal cord and sacral agenesis. Open NTDs are more common than closed NTDs.[4]

Anencephaly and spina bifida are account for up to 95% of all NTDs.[5] The incidence of meningocele is 5% while myelomeningocele is 90-95% in all spina bifida cases, which is the most severe birth defect compatible with survival.[6,4,7] About 70-80% of encephaloceles occur in the occipital region and the nasal and parietal ones are less common.[9]

The presentation of spina bifida is usually with fluid filled sac commonly at the lumbosacral region (45-50%), but it can be at any site from cervical to sacral region.[6,7]

Individuals with spina bifida have substantially enhanced survival rate due to recent improvements in medical and surgical management. However, these patients continue to be at increased risk for morbidity and mortality throughout their life. These patients experience varying degrees of motor and sensory dysfunction of lower limbs and failure of anal and urethral sphincters.[9]

The incidence of hydrocephalus associated with myelomeningocele ranges from 80-95%.[4] Other associated anomalies include skeletal abnormalities (i.e. kyphosis, scoliosis), genito urinary anomalies (20% cases) (renal cysts, duplication of genitourinary tract and undescended testes), Chiari II malformation (>90%), club foot and congenital heart disease (VSD).[7,4,1] In spite of numerous progresses that have been done towards understanding the causation of the majority of the cases still unknown. Certain epidemiologic genetic studies have suggested some high risk groups. These include maternal age of <20y or >35y, a past history of having a fetus with NTDs, poor socioeconomic status, gross nutritional deficiency (folic acid), two extremes of parity (primipara and grand multipara) and lack of proper ANC.[3,10] Adequate Folic acid consumption during early pregnancy prevents the occurrence and recurrence risk of NTDs by 50%-70%.[11,4] Paternal occupational factors like agriculture, painting, welding have also been linked with increased risk of NTDs.[3]

Association of genetic factors is also advocated in many cases. The risk of 1st child having spinal abnormality is around 0.1%-0.2%. But having one affected sibling the risk of a 2nd child is 2%- 5%, and the risk of 3rd affected child is compounded to 10%-15%.[3] Numerous teratogens have been implicated in the etiology of NTDs, like radiations, hyperthermia, heat exposure and hot-tub, hypervitaminosis A, viral infections, drugs like carbamazepine, valproic acid and use of folic acid antagonists (methotrexate).[3,5]

The severity of NTDs depends on the level and degree of spinal cord exposure. The symptoms are ranging from mild cosmetic symptoms to severe paralysis, chronic infections, incontinence or retention. Patients are also at increased risk of developing hydrocephalus that requires ventriculo-peritoneal (VP) shunts.[11]

The prognosis is also dependent to the level of the lesion. So, defining of lesion localization with maternal ultrasonography at mid-trimester is useful to predict the short and long term outcomes.[12]

The standard of care includes diagnosis in utero by proper ANC, proper counseling of the parents, caesarian section rather than vaginal delivery, surgical closure as early as possible. Continued monitoring for the development of hydrocephalus is necessary, with surgical intervention as needed.[11]

In the management of myelomeningocele, the patient without overt clinical hydrocephalus, most surgeons used to repair the myelomeningocele only. But that may result subsequent hydrocephalus in 65 to 85% within 6 months and also can cause breakdown of repair and CSF leakage. To prevent this complication some surgeons begins to do repair of myelomeningocele and introduction of VP shunt at least 3 days after the initial repair. This gives better result. But also increase hospital stays and imposes an additional operative trauma within a short interval. It also increases the incidences of wound related complications. Significant improvement has been shown in the management of myelomeningocele by practicing simultaneous placement of VP shunt during repair of myelomeningocele. This decreases the hospital stay, prevents failure of the repair, prevents or delays the development of subsequent hydrocephalus.[13]

**Objectives:**

*General objective:*

To observe the outcome of surgical management of the neural tube defects (NTDs) in children.

*Specific objective:*

To record the different types of NTDs, to evaluate the different types of post-operative complications, to manage the different modalities of post-operative complications, to observe the overall surgical outcome of NTDs.

**Methodology:**

This prospective observational study was conducted in the Department of Paediatric Surgery, Sylhet M.A.G. Osmani Medical College & Hospital during the period from March, 2018 to August, 2020. Thirty children with a neural tube defect during the study period were included. Associated with life threatening major congenital malformations were excluded.

**Study Procedure**

All the admitted patients with NTDs has been assessed. Patient has been advised for CBC, BT, CT, Blood grouping and Rh typing, Urine R/M/E, X-ray lumbosacral spine both A/P and lateral view, Chest x-ray P/A view, USG of the lesion, USG of brain, USG of whole abdomen and 2D echocardiography to detect the extent of the disease, associated anomalies and to prepare the patient for surgical management needed according to types. Closure includes partial excision of the sac, water tight closure of the dura and reconstruction. Tension-free closure of the skin has been ensured after keeping a tube drain in situ. Most of the patients with NTDs were operated ≤7 days of admission. The follow-up schedules were on 15th POD, at monthly upto 6 months and at 1 year after operation. In the follow-up we looked for CSF leakage from the wound and wound infection, paralysis of lower limbs or club foot, incontinence of urine and faeces, hydrocephalus. The prognosis was recorded by taking detailed history from the parents and proper physical examination of the patients.

**Statistical analysis**

Data were processed manually and analyzed by the help of Microsoft Office Excel and statistical package for social sciences (SPSS) version 25.

Qualitative data were presented as frequency and percentage; and quantitative data were presented as mean and standard deviation.

Chi-square ($\div^2$) test was done to find out the level of significance between two categorical variables. A probability value (p) of <0.05 was considered statistically significant.

***Ethical consideration***

An approval of the study protocol was obtained from the institutional Ethical Review Committee of Sylhet M.A.G. Osmani Medical College, Sylhet before the commencement of the study.

**Operational definition:**

**Recovery without complication:** Recovery from primary pathology without complication.

**Recovery with complication:** Recovery from primary pathology with some complications.

**Results:**

**Table 1 :** *Showed the distribution of the patients on the basis of age group.*

| Age | Number | % |
|---|---|---|
| 1-7 days | 20 | 66.7 |
| 8-30 days | 4 | 13.3 |
| <30 days | 6 | 20.0 |

**Table 2:** *Showed the frequency distribution of patients according to gender.*

| Gender | Number | % |
|---|---|---|
| Male | 21 | 70.0 |
| Female | 9 | 30.0 |

**Table 3:** Showed the gestational age of the patients. Preterm birth was in 6 (20.0%) patients, who were associated with recovery with complications and morality.

| Gestational age | Number | % |
|---|---|---|
| Term | 24 | 80.0 |
| Preterm | 6 | 20.0 |

**Table IV** *showed the periconceptional folic acid supplementation in NTD.*

| Folic acid supplementation | No. | % |
|---|---|---|
| Yes | 3 | 10.0 |
| No | 27 | 90.0 |

**Table V** *Showed the distribution of the patients according to type of neural tube defects. Patients with myelomeningocele have more associated birth defects.*

| Type of NTDs | Number | % |
|---|---|---|
| Meningocele | 2 | 6.7 |
| Myelomeningocele | 22 | 73.3 |
| Encephalocele | 3 | 10.0 |
| Lipomeningocele | 3 | 10.0 |

**Table VI:** *Showed the distribution of the patients according to the site of neural tube defects. The patients who had been developed postoperative complications, the defect was presented in the lumbosacral region.*

| Site of NTDs | Number | % |
|---|---|---|
| Lumbosacral | 23 | 76.7 |
| Sacral | 2 | 6.7 |
| Occipital | 2 | 10.0 |
| Cervical | 1 | 3.3 |
| Lumbar | 1 | 3.3 |

**Table VII** *Revealed that the nature of neural tube defects was ruptured in 18 (60.0%), among them 7 (23.3%) developed post-operative complications. Morbidity and mortality was more in ruptured cases.*

| Nature of NTDs | Number | % |
|---|---|---|
| Ruptured | 18 | 60.0 |
| Not-ruptured | 12 | 40.0 |

**Table VIII** *Associated congenital defects*

| Associated congenital defects | Number | % |
|---|---|---|
| Hydrocephalus | 17 | 56.7 |
| Club foot | 5 | 16.7 |
| Cervical prolapse | 2 | 6.7 |
| Kyphosis | 2 | 6.7 |
| No | 12 | 40.0 |

**Table IX** *Associated neurological defects*

| Associated neurological defects | Number | % |
|---|---|---|
| Lower limb paresis/paralysis | 25 | 83.3 |
| Urinary incontinence | 15 | 50.0 |
| Faecal incontinence | 12 | 40.0 |
| No | 5 | 16.7 |

**Table X** Age of child during surgical management

| Age | Number | % |
|---|---|---|
| ≤ 7 days | 20 | 66.6 |
| >7 days | 10 | 33.3 |

**Table XI** *Postoperative complications*

| Postoperative complications | Number | % |
|---|---|---|
| Wound infection | 3 | 10.0 |
| Wound dehiscence | 5 | 16.7 |
| CSF leakage | 7 | 23.3 |
| Meningitis | 3 | 10.0 |
| No | 20 | 66.7 |

**Table XII** Treatment of postoperative complications

| Treatment of postoperative complications | Number | % |
|---|---|---|
| Conservative | | |
| Antibiotics | 10 | 33.3 |
| Regular dressing | 8 | 26.7 |
| Surgical | | |
| Secondary closure | 6 | 20.0 |

**Table XIII** Treatment of associated anomalies

| Treatment of associated anomalies | Number | % |
|---|---|---|
| V-P shunt | 12 | 40.0 |
| Physiotherapy | 25 | 83.3 |
| Other | 4 | 13.3 |
| No | 5 | 16.7 |

**Table XIV** *Duration of hospital stay*

| Duration of hospital stay | Number | % |
|---|---|---|
| ≤ 10 days | 20 | 66.7 |
| >10 days | 10 | 33.3 |

**Table XV** *Follow up at 15 days after operation*

| Outcome | Number | % |
|---|---|---|
| Wound infection | 3 | 10.0 |
| Lower limb paresis/paralysis | 23 | 76.7 |
| Faecal incontinence | 10 | 33.3 |
| Urinary incontinence | 13 | 43.3 |
| Hydrocephalus | 15 | 50.0 |

**Table XVI** *Follow up at 1months, 3 months, 6months and 12 months after operation*

| Outcome | 1m | 3m | 6m | 12m |
|---|---|---|---|---|
| Lower limb paresis/ paralysis | 76.7% | 76.7% | 76.7% | 70.0% |
| Faecal incontinence | 33.3% | 33.3% | 33.3% | 33.3% |
| Urinary incontinence | 43.3% | 43.3% | 43.3% | 43.3% |
| Hydrocephalus | 60.0% | 60.0% | 60.0% | 60.0% |

**Table 17** *Outcome (n=30)*

| Outcome | Number | % |
|---|---|---|
| Recovery without complication | 20 | 66.7 |
| Recovery with complications | 8 | 26.7 |
| Death | 2 | 6.7 |

**Discussion:**

Goswami et al., (2015) found that age at presentation of neural tube defect ranged from 1 day to 5 years. In this study the ages of the patients ranged from 1day to 9 years with the median age of 3.5 days. There were 20 (66.7%) patients in the age group of 1-7 days, 4 (13.3%) patients in the age group of 8-30 days and 6 (20.0%) patients in the age group of above one month. Delaying in presentation yet might be due to delayed referral, financial problems and sometimes folk cultural and religious beliefs, that NTDs are a punishment from God. This study showed that there were 21 (70.0%) male and 9 (30.0%) female with a ratio of male to female was 2.33:1. Tendency to neural tube defect was more frequent in male than that of female (p=0.028). Rai et al., (2016) found that the frequencies of a male child affected to that of the female child were more with a ratio of 1.38:1 in neural tube defects. Many studies revealed that females are more affected than males.[5, 18]

This study showed that preterm birth in 6 (20.0%) patients and term birth in 24 (80.0%) patients of NTD. But Gedefaw et al., (2018) found preterm birth was more common in NTD (69.4%). In another study Yin et al., (2020) also found preterm birth was more common in NTD (84.5%).

It is mostly accepted that inadequate intake of natural folate, or its synthetic form folic acid, before and during early pregnancy is associated with increased risk of spina bifida.[20,21] In this regards we found most of the mother of the patients did not take periconceptional folic acid supplementation (90.0%) and only 10.0% of mother of the patients take periconceptional folic acid supplementation; difference was statistically significant (p<0.001). Motah et al., (2017) found none of the mothers of baby with NTDs in their series was supplemented for folic acid before pregnancy and most of them who took folic acid did not do so during the critical period. Alatise et al., (2006) reported that only 3 mothers in their series took folic acid before pregnancy. This might have been due to several reasons; lack of experience, and most of the time, the mothers were not even knowing that they were pregnant during the critical period.

In our study myelomeningocele (73.3%) was the most common neural tube defects and others were encephalocele (10.0%), lipomeningocele (10.0%) and meningocele (6.7%). Gedefaw et al., (2018) found that 54.1% were cases of anencephaly; 40.5%, spina bifida; and 5.4% encephalocele. The frequency of spina bifida was in higher proportion compared to anencephaly reported in the study of Rai et al., (2016). However, many studies have reported equal incidences of these two forms of NTD.[22, 23]

The site of neural tube defects was lumbosacral in 23 (76.7%) cases, occipital in 3 (10.0%) cases, sacral in 2 (6.7%) cases, lumber in 1 (3.3%) cases and cervical in 1 (3.3%) cases. Motah et al., (2017) found the presenting sites for NTDs at lumbosacral in 47.7% of cases, lumber in 41.3% of cases, sacral in 6.6% of cases, and thoracolumbar in 4.3% of cases.

This study revealed that the nature of neural tube defects was ruptured in 18 (60.0%) and not ruptured in 12 (40.0%) cases. This result was consistent with the study of Motah et al., (2017); where neural tube defects were ruptured in 52.2% of cases and un-ruptured in 47.8% of cases.

Associated congenital defects in this study were hydrocephalus in 56.7%, club foot in 16.7%, cervical prolapse in 6.7% and kyphosis in 6.7% cases. Motah et al., (2017) also found associated birth defects were

hydrocephalus (65.3%), bilateral talipes calcaneovalgus (18.4%), bilateral talipes equinovarus (16.3%) and several other less frequent defects.

Associated neurological defects were lower limb paresis/ paralysis in 83.3%, Urinary incontinence in 50.0% and faecal incontinence in 40.0% of cases. Motah et al., (2017) reported neurological defects associated with NTD were lower limb paresis/ paralysis in 65.33%; and urinary incontinence and faecal incontinence deficit in 61.2% of cases Kumar and Singh, (2003) and Alatise et al., (2006) had been reported similar results. Sphincteric dysfunctions were more frequent in patients presented with myelomeningocele.

The definitive procedure for NTDs is operative closure of the defect within. Closure includes partial excision of the sac, water tight closure of the dura mater and reconstruction. None of the patient in this study had been operated within 72hrs of age. The ages of the child during operation are d"7 days for 20 (66.7%) patients and >7 days for 10 (33.3%) patients.

Postoperative complications in this study were wound infection in 10.0%, wound dehiscence in 16.7%, CSF leakage in 23.3% and meningitis in 10.0% cases. Treatment of postoperative complications was administration of antibiotics (33.3%), regular dressing (26.7%) and physiotherapy (10.0%) for cases as conservative treatment; and 6 (20.0%) patients required surgical management (e.g. secondary closure). The 10 (33.3%) patients who had developed postoperative complications were operated after 7 days of age, and the defect was presented in the lumbosacral region. Among them, myelomeningocele was in 7 (23.3%) patients and lipomeningocele was in 3 (10%) patients. The overlying skin was ruptured in 7 (23.3%) patients and not ruptured in 3 (10%) patients. Motah et al., (2017) observed patients with complications following surgery were 55.1%. The most common post-operative complication was wound infections (44.9%). Others were wound dehiscence (40.8%), CSF leakage (20.4%) and meningitis/sepsis (32.7%).

**Conclusion:**

Neural tube defects (NTDs) are more common in male child in this study. Myelomeningocele is the most common type and lumbo-sacral was the most frequent clinical type of neural tube defects (NTDs). Associated common birth defect is hydrocephalus and neurological defect is lower limb paresis/ paralysis. Recovery without complication is more frequent than recovery with complications and death. Early operation is associated with recovery without complication and less morbidity.

**Limitations of the study:**

This study was not without limitations. The limitations of the study were-

None of the patient have been operated before 72hrs of life, lack of opportunity for MRI facility, lack of expert neonatal neurosurgeon, lack of dedicated neonatal anaesthesiologist, several surgeons performed the operation, lack of opportunity for NICU support, lack of multidisciplinary approach, follow up period was short.

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