International Journal of Life Sciences, Biotechnology and Pharma Research Vol. 14, No. 4, April 2025 Online ISSN: 2250-3137
Print ISSN: 2977-0122

DOI: 10.69605/ijlbpr_14.4.2025.4

ORIGINAL RESEARCH

Peri - operative complications in paediatric patients undergoing surgery for neural tube defects- an observational study

[1]Kavipriya S, [2]Anju Paul, [3]Anita Shetty, [4]Uma Kamat, [5]Sangeetha B S, [6]Chaitra N H

[1]Senior Resident, [4]Assistant Professor, Department of Anaesthesiology and Critical Care, Seth GSMC and KEM Hospital, Mumbai, India
[2]Assistant Professor, Department of Anaesthesiology and Critical Care, Government Medical College, Jalgaon, India
[3]Professor & Head of Department, Department of Anaesthesiology and Critical Care, HBT Medical College and R.N.Cooper Municipal General Hospital, Mumbai, India
[5]Senior Resident, Department of Anaesthesiology and Critical Care, Grant Government Medical College and J.J. Hospital, Mumbai, India
[6]Senior Resident, Department of Anaesthesiology and Critical Care, KB Bhabha Municipal General Hospital, Mumbai, India

Corresponding Author
Anju Paul
Assistant Professor, Department of Anaesthesiology and Critical Care, Government Medical College, Jalgaon, India
Email: anjupaul2@gmail.com

Received: 26 February, 2025 Accepted: 20 March, 2025 Published: 31 March, 2025

ABSTRACT

Background: Neural tube defects(NTD) are congenital birth defects of central nervous system, caused by failure of neural tube closure during embryogenesis. Aetiology of NTD is considered to be multifactorial [1] and Incidence is 0.5 to >10 per 1000 pregnancies worldwide. NTD have various associated neurological abnormalities and systemic anomalies. Ultimately, they pose challenges to anaesthesiologists because of difficult airway and perioperative complications, attributable to their age group, co-morbid conditions and associated anomalies.[2] Henceforth,thorough knowledge of perioperative complications are needed for a meticulous anaesthetic planand prevention of further neurological damage and better quality of life.[3] **Aim and Objectives:** To observe the anaesthetic challenges and perioperative complications in patients undergoing NTD repair. **Materials and Methods:** We had conducted an observational study **both Retrospective and Prospectively** on the children operated for NTD **(Meningomyelocele, Encephalocele and Tethered cord)** for a duration of 2 years from June 2019-May 2021. Data regarding their Pre anaesthetic assessment, Intra-operative management, Perioperative complications were collected and analysed. **Results:** Out of 78 patients with neural tube defects, 34 were males(43.5%) and 44 females(56.4%). Meningomyelocele (71.7%) was the highest among all three defects. Lumbosacral region(44.8%) was the most common site. Most associated abnormalities were Hydrocephalus(16.6%) and Arnold Chiari malformation(9.0%). Pre-op complications include complications associated with lesions,associated congenital multisystem abnormalities and coexisting medical disorders. Intra-operatively, Cardiovascular complications such as hypotension (10.2%) bradycardia(6.4%) and tachycardia, hypoglycaemia,hypothermia and cervical cord handling and related complications were noted.Cardiovascular complications are more common than respiratory ones. Post-operatively, Three(3.9%) had complications and Four(5.1%) required ventilator support. **Conclusion:** Children with NTD are vulnerable for peri-operative complications. So, Meticulous anaesthetic approach with Early surgical repair is required for good prognosis.

Keywords: Anaesthesia, Neural Tube Defects, Meningomyelocele, Encephalocele

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INTRODUCTION

Neural tube defects (NTD) occur because of failure in neural tube closure during embryogenesis. They can be open or closed. And may arise anywhere along the neural axis from head to spine resulting in Anencephaly or Craniorachischisis, Spina Bifida respectively. Types of Spina Bifida are Meningocele, Meningomyelocele and Myelocele. Generally, these are open defects.NTD(almost 2/3rd meningomyelocele) are associated with neurological abnormalities such as Chiari malformation, hydrocephalus, developmental brain defects leading to

DOI: 10.69605/ijlbpr_14.4.2025.4

neurological deficits, bowel and bladder incontinence.[2] Also, there are incidences of various other anomalies like congenital heart diseases, orthopaedic and genitourinary disorders, thereby making these children prone for severe morbidities and lifelong disabilities.[4]So knowledge of perioperative complications are needed for meticulous anaesthesia plan to prevent further complication. Here we have done an observational study to find out common perioperative complications and anaesthetic challenges in patients posted for repair of neural tube defect in Indian population.

METHODOLOGY

This study was conducted as an observational study both Retrospective and Prospectively on the children operated for NTD (Meningomyelocele, Encephalocele and Tethered cord**)** for a duration of 2 years from June 2019-May 2021in the tertiary care centre in India after obtaining ethical committee approval EC/154/2019. All the patients of paediatric age group (birth - less than 12 years) who presented with 1) Meningomyelocele, 2) Encephalocele and 3) Tethered cord for Repair were included in the study.Parent or Guardian of the child, who was not willing to give consent for the study were excluded.

SAMPLE SIZE CALCULATION

A study conducted by Chand MB et al, inferred, the least common complication perioperatively was Endobronchial intubation and the prevalence of which was 5.4%. Using this with 95% confidence interval and 5% absolute error, we calculated the minimum sample size to be 78.

Following formula has been used to calculate the sample size

$SS = (1.96)^2 p (1-p)/r^2$

SS - sample size

p - prevalence of the complication

r - absolute error

Henceforth for the study, we needed to enrol a minimum 78 patients who had undergone NTD repair.All the available medical records of children who had undergone repair of above mentioned NTD, were analysed for Retrospective study starting from June 2019 till ethics committee clearance. Detailed analysis regarding their Pre anaesthetic evaluation, Intra-operative course and Post-operative complications was carried out.Informed written consent from parent or guardian and Assent for children aged over 7years, Prospective observational study was conducted till May 2021, thus altogether for a total duration of two years.

Pre-operative assessment comprised of Demographic details (age, sex, height, weight) detailed history, general and systemic examination (for general condition, neurological status and associated systemic abnormalities)examination of lesion (site, extent and CSF leakage)and basic investigations (Hemogram, serum electrolytes), Radiological investigations (to know extent of lesion) and other relevant tests pertaining to the case were done and optimised.On the day of surgery after confirming adequate hours of starvation, child was taken into operation theatre and monitoring of basic parameters were started which includes Heart rate, Blood pressure, Oxygen saturation, Capnography, Urine output and Temperature. Routine induction using Inj.Fentanyl (2mcg/kg) for analgesia and Inj.Propofol (2mg/kg) were used. Inj.Atracurium (0.05mg/kg) was used for neuromuscular blockade.Intubation was performed in supine position, with stack of head rings, bolster under the shoulders and doughnut below the lesion (inorder to prevent the damage of neural structures). Anaesthesia was maintained with oxygen, nitrous oxide and sevoflurane plus controlled ventilation. Dextrose with Ringer Lactate (DRL) was the fluid of choice in most patients. **Intra-operative** monitoring included Anaesthetic technique, airway management with difficulties encountered, fluids infused and blood loss. At the end of surgery, neuromuscular blockade was reversed withInj.neostigmine(0.05mg/kg) and Inj.glycopyrrolate(10mcg/kg). Patients were extubated when they are fully awake with intact reflexes and patients who were not in a condition to extubate were kept on mechanical ventilation.Preop, intraop and post-op complications were recorded.Data regarding their Pre anaesthetic assessment, Intra-operative management, Perioperative complications were collected and entered in a Microsoft excel sheet and analysed using **SPSS version 21.** Descriptive statistics were used for analysis.Quantitative variables were represented as Mean±SD, or median (interquartile range). Qualitative variables were represented as frequencies and percentages.

RESULTS

In our study out of 78 patients with neural tube defect,34 patients were male and 44 were females.Female: male ratio is 1.2:1 (chart 1).Among all three NTD**Meningomyelocele was the most common defect(56 cases,71.7%)** (chart 2).Out of which, 8 of them(14.2%) also had Tethered cord.Considering all three NTD, Lumbosacral region(44.8%) was the most common site of NTD and Lumbar region(19.2%) was the second common site.(Table 1)

Preoperative complications because of lesions are in the of Recurrence, Ruptured lesions with CSF leak, Neurological Deficit and Bowel/Bladder dysfunction. (Table 2)Ruptured lesions with CSF leak is the most common one (19.2%). There are multiple associated system abnormalities were noted. Hydrocephalus(16.6%), Arnold Chiari malformation type 2 (7.7%) and Club foot(5.1%) were the most common abnormalities associated with NTD.(Table 3) Children posted for NTD repair had multiple co-existing medical problems. Among these **Anaemia was the most common (37.1%)** followed by **Hyponatremia(28.2%)** (Table 4).Other abnormalities

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DOI: 10.69605/ijlbpr_14.4.2025.4

showed in table5. Most common age group operated operated for neural tube defect was **0-60days (56.4%)** (chart 3). All babies were intubated in the supine position. The averageduration of surgery is 3.5 hours. Intra-operative an average blood loss was 80ml.Average CSF loss was in the range of 40-215ml. Blood and blood products were transfused in an average of 60ml.

Intra-operatively, cardiovascular complications were more common than respiratory complications.(Table 6).The most common was Hypotension followed by Bradycardia. The cause of hypotension in 50% of patients was blood and CSF loss, corrected by fluids and blood transfusion. Inotrope support was required in one case, which was slowly tapered and stopped once after patient became hemodynamically stable. The cause of bradycardia in 2cases was hypothermia, reverted with forced air warming and warm fluids. There were no Respiratory Complications in our study.Cervical cord handlinghappenedin one case of cervical meningomyelocele which cord edema and apnoeic/ episodes and the the child was continued on mechanical ventilation post-operatively.

Out of 78cases, 74 had smooth recovery. 4 were shifted on ventilator support(3 due to inadequate respiratory efforts and 1 due to risks of cord edema) (chart 4). Out of those, **two** were extubated on the same day. Other two were continued on mechanical ventilation.

Post-operative complications occurred in 3 cases(3.9%). Of these three, one had wound infection for which wound debridement with right VP shunt insertion was done. Another one had CSF leak, for which re-exploration was done. One succumbed due to cardio respiratory arrest on first post-operative day due to cervical cord edema (chart 5).

During post-operative period, only one casehad improvement of neurological deficit and one had worsening of deficit. The rest of the cases showed no improvement. Out of 78, 15children had History of NICU admission mostly due to respiratory distress, HIE, NTD and jaundice.Apart from excision and repair, additional procedureslike skin grafting, flap cover, simultaneous detethering of cord and ventriculo peritoneal shunting were required in 14 cases of meningomyelocele(18%).

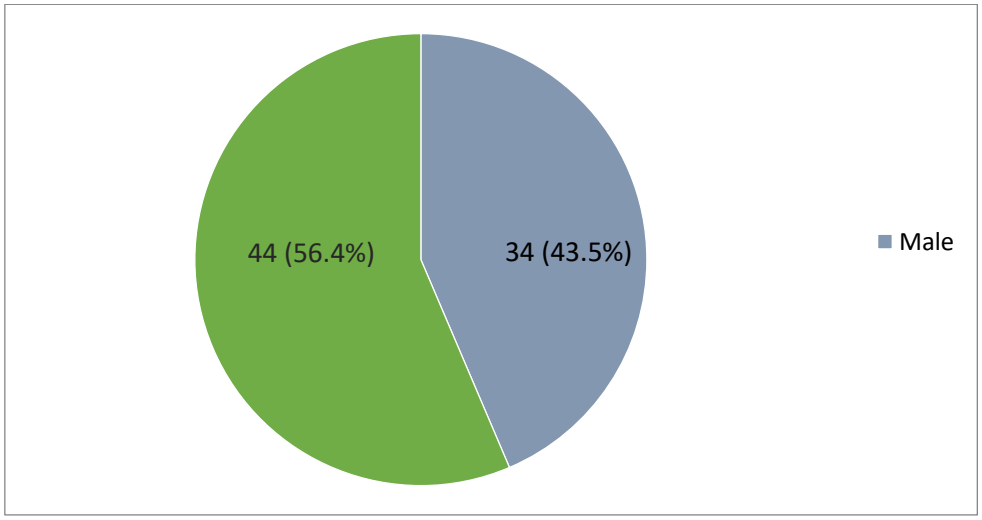

Chart 1: Sex of the child

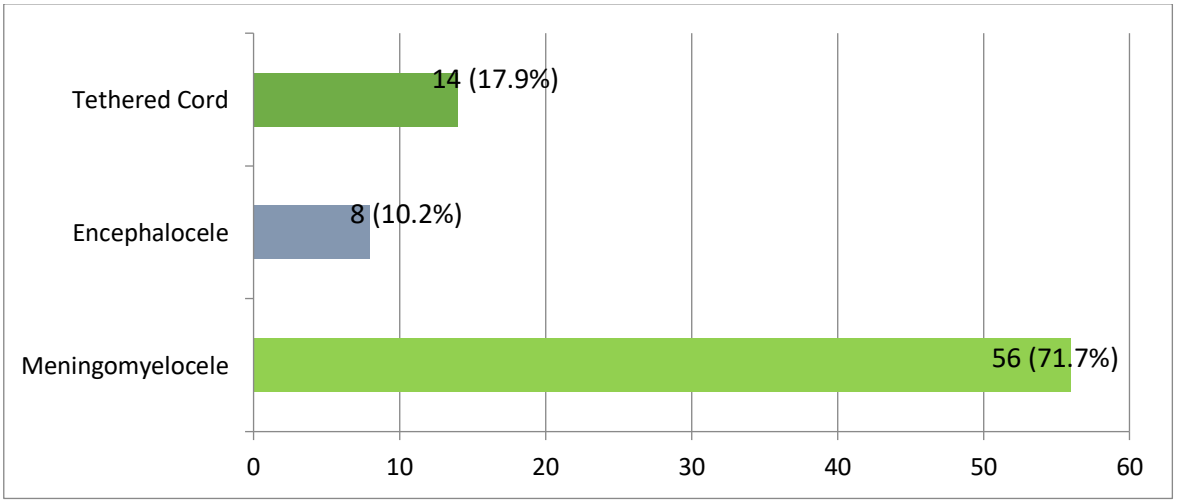

Chart 2: Type of Defect

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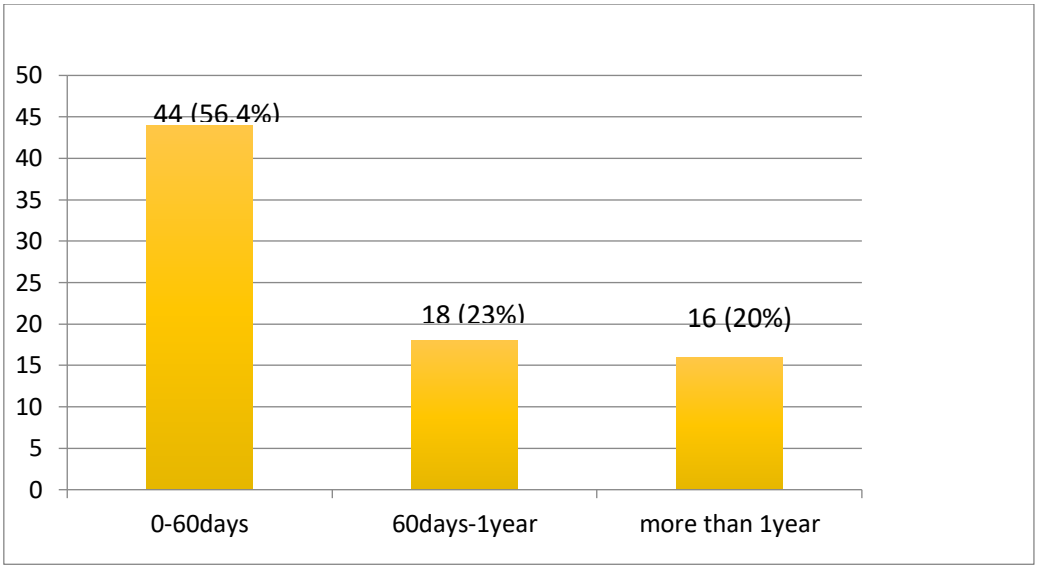

Chart 3: Age of the child

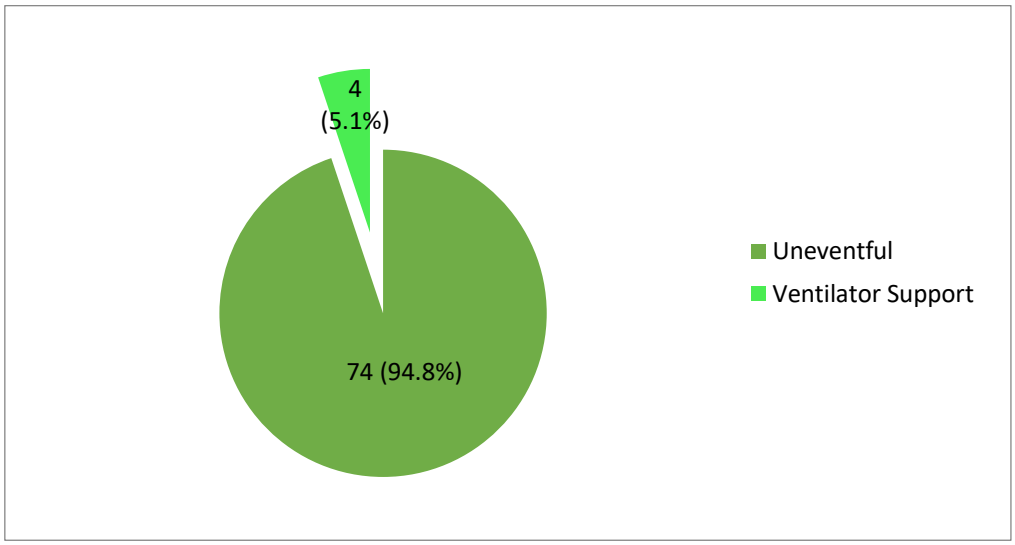

Chart 4: Post operative Recovery

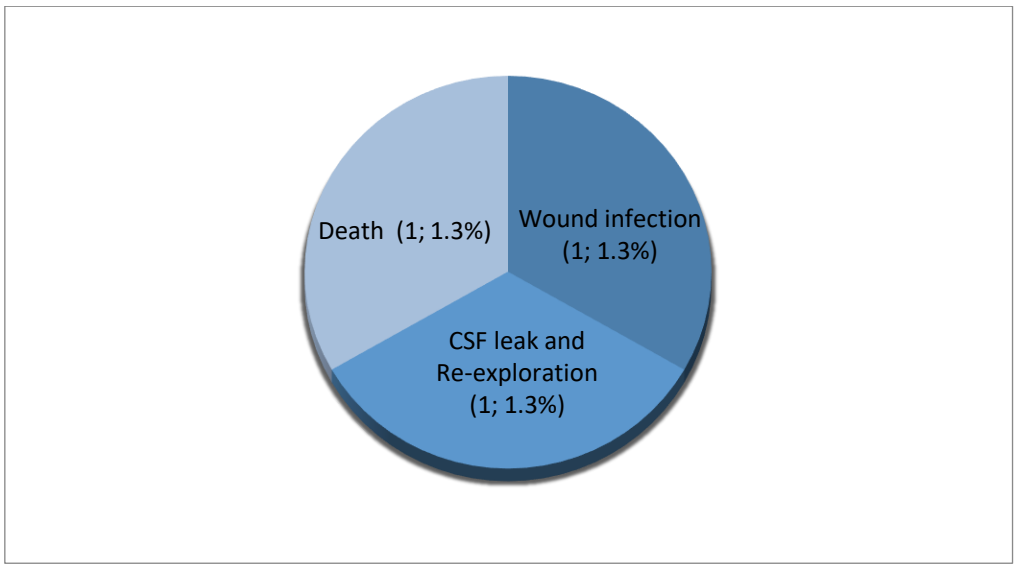

Chart 5: Post-operative Complications

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Table 1: site of lesion

Site of lesion	Meningomyelocele	Encephalocele	Tethered cord	Total
Lumbosacral	**29 (51.7%)**	-	6 (42.9%)	35 (44.8%)
Lumbar	7 (12.5%)	-	**8 (57.1%)**	15 (19.2%)
Thoracolumbar	12 (21.4%)	-	-	12 (15.3%)
Thoracic / dorsal	3 (5.35%)	-	-	3 (3.84%)
Sacral	4 (7.14%)	-	-	4 (5.1%)
Occipital	-	**6 (75%)**	-	6 (7.7%)
Cervical	2 (3.6%)	-	-	2 (2.5%)
Frontonasal	-	2 (25%)	-	2 (2.5%)
Total	**56**	**8**	**14**	

Table 2: Pre-operative complications due to lesion

Complications associated	Frequency	Percentage
Recurrence	4	5.1
Ruptured lesions with CSF leak	15	19.2
Neurological Deficit	18	23
Bowel/Bladder dysfunction	3	3.8

Table 3: Associated abnormalities

System	Associated anomaly	Frequency	Percentage
Central nervous system	Hydrocephalus	13	16.6
	Arnold chiari malformation type 2	6	7.7
	Chiari malformation type 3	1	1.3
	Syringomyelia	3	3.8
Musculoskeletal system	Scoliosis	3	3.8
	Clubfoot	4	5.1
	Tropic ulcer	1	1.3
	Absent ribs(left side)	1	1.3
Genitourinary system	Hydronephrosis	1	1.3
	Agenesis of left kidney	1	1.3
Cardiovascular system	Atrial Septal Defect	1	1.3
	Patent Foramen Ovale	2	2.5

Table 4: Coexisting medical disorders

Coexisting medical disorders	Upper respiratory tract infection	5	6.4
	Cystitis	1	1.3
	Meningitis	1	1.3
	Anaemia	29	37.1
	Electrolyte disturbances: a.Hyponatremia	22	28.2
	b.Hyperkalemia	4	5.1
	c.Hypokalemia	3	3.8

Table 5: Other significant abnormalities

Abnormal findings	Frequency	Percentage	Type of defect present
Periorbital swelling (bilateral)	1	1.3	Encephalocele
Squint	1	1.3	Encephalocele
Dermal sinus	2	2.5	Meningomyelocele
Tuft of hair (sacral area)	1	1.3	Tethered cord

Table 6: Intra-operative Complications

Complications		Frequency	Percentage
Cardiovascular	Bradycardia	5	6.4
	Tachycardia	1	1.3
	Hypotension	8	10.2
	Hypertension	NIL	NIL
	Hypoxemia		

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Respiratory	Bronchospasm	NIL	NIL
	Laryngospasm		
	Endobronchial intubation		
	Accidental extubation		
Other Complications	Hypothermia	2	2.5
	Hypoglycaemia	3	3.8
	Cervical cord handling & Risk of apnoea	1	1.3

DISCUSSION

Neural tube defects(NTD) are the spectrum of congenital anomalies including cranial defects and spinal dysraphisms. These can be open or closed. Female children are commonly affected in our study. Similarly, **Elmesallamy et al** studyon 43 patients for tethered cord syndrome repair, showed more females were operated.[5] The aetiology is multifactorial and occurs due to the interaction of multiple maternal, genetic and environmental factors.[6] Out of all three NTD in our study, meningomyelocele was the most common defect(71.7%) followed by tethered cord(17.9%). Least common was encephalocele(10.2%).

Clinical presentations of NTD vary from developmental delay, neurological deficit to sphincter dysfunction (bowel or bladder). Neurological deficits are more common in meningomyelocele. Patients with cervical lesions present with quadriplegia and thoracic or lumbosacral lesions present with paraplegia.[7] In our study, out of three cases of neurogenic bladder, two(66.6%) were found inpatients with tethered cord.**Elmesallamy et al.** found that all patients in their study with Tethered cord had neurogenic bladder [5]

Several neurological abnormalities such as Arnold chiari malformation, hydrocephalus and syringomyelia are commonly associated with NTD Developmental disorders of brain such as microgyria, corpus callosum agenesis, arachnoid cyst and porencephalic cyst are common findings apart from Chiari malformations.[8] hydrocephalus will necessitate a drainage procedure to be performed—either a ventriculo-peritoneal shunt or a ventriculostomy.[2]. Due to brainstem involvement in Arnold Chiari malformations, there are chances of respiratory distress, apnoea, bradycardia, dysphagia with pulmonary aspiration, torticollis, nystagmus, hypotonia, spasticity, cranial nerve dysfunction and inspiratory stridor due to vocal cord paralysis.[9]During laryngoscopy and intubation,extreme head flexion or extension may cause brainstem compression resulting in bradycardia and even cardiac arrest.[10] These children also have abnormal responses to hypoxia, hypercarbia due to cranial nerve and brainstem dysfunction (Ward et al. 1986).Hydrocephalus contributes to difficult airway, abnormal response to hypoxia and susceptibility for post-operative apnea**.[8]**It also influences the neurocognitive outcome and increases the morbidity of the child if there is shunt malfunction or infection.[10]

In our study, hydrocephalus was the most common neurological abnormality found in 13 children(16.6%) and then secondly Chiari malformation typeII(7.7%).In **Chand MB et al.** study on37children undergoing meningomyelocele repair, similarly revealed hydrocephalus to be the most common neurological abnormality (67.56%).[11]

Other congenital anomalies like cardiovascular defects, genitourinary disorders, orthopaedic deformities might be present in NTD, as they develop concurrently with deformed neurological system during embryogenesis.[7] In our study, Clubfoot was found in four(5.1%) and Scoliosis in three(3.8%) patients of meningomyelocele. Similarly, **Chand MB et al.** noted scoliosis in 12cases of meningomyelocele(32%) and clubfoot in 11cases(29%).[11]

Children with NTD are also vulnerable to medical disorders. Most common medical disorder was anaemia(37.1%) followed by hyponatremia(28.2%). Significantly, they were noticed in meningomyelocele(72.3%) followed by tethered cord(15.3%). **Ravindra S Giri et al.** reported anaemia in 2 cases(10%), electrolyte imbalances in four(20%) and upper respiratory tract infection in four(20%) cases.[12]

Thorough **pre-operative evaluation** of NTD plays a crucial role. According to literature, 37% of meningomyelocele is associated with congenital heart disease and 36% associated with short trachea, which might lead to endobronchial intubation.[9] In ours, we didn't have any endobronchial intubation.**Early surgical repair** of neural tube defects is essential **within 24-48 hours** as it reduces the infection, improves the survival rate, enhances the neurological outcome and long-term prognosis. **Prenatal surgery** has been proven to be more effective in reducing future complications.[13]The major age group operated were 0-60 days old(56.4%). **Chand MB et al and Ravindra S Giri et al.** studies on meningomyelocele and encephalocele, noted more children aged 60 days-1year for repair.[12,11]

Out of all three, meningomyelocele and encephalocele require **excision and repair** whereas, tethered cord needs complete **detethering of cord via release of filum terminale**. Sometimes, additional procedures such as shunt insertion, posterior fossa decompression would be required in symptomatic cases.[14] In our study,14(18%) required shunt insertion.

During all these procedures, strict antiseptic techniques should be followed under proper antibiotic

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DOI: 10.69605/ijlbpr_14.4.2025.4

cover. **Latex free sterile gloves should be used,** as these children are prone for latex allergy.[15]

Anaesthesia induction can be Intravenous or Inhalational. A major anaesthetic consideration is **positioning of the neonate** during induction. It is essential to have **no direct pressure on the defect**.[6]

Other concerns in airway management are **Difficult intubation** attributed by hydrocephalus, larger occipital or cervical lesions and anatomical differences in the paediatric airway. **Difficult bag mask ventilation** in frontonasal encephaloceles. Rise in Intracranial pressure due to mask ventilation or intubation.

Peri-operative complications in the NTD repair always pose challenges for anaesthesiologists. Respiratory complications are bronchospasm, laryngospasm, hypoxemia, apnoeic episodes, accidental extubation(due to prone position), endobronchial intubation
(due to short trachea). Thecardiovascular complications are bradycardia, tachycardia, hypotension and cardiac arrest rarely. **Bradycardia** most commonly due to brainstem compression or vagal stimulation during laryngoscopy or surgery. **Hypotension** due to blood loss or sudden CSF loss resulting in brain herniation.[10] In our study, Cardiovascular complications (Hypotension 10.2% and Bradycardia 6.4%) were common than respiratory ones. Bradycardia was partly due to Hypothermia. **Ravindra S. Giri et al.** study showed respiratory complications (45%) were common than cardiovascular ones.[12]

Other uncommon complications include hypothermia, dislodgement of intravenous catheters, facial edemaand bleeding due to prone positioning..[16]

Hypothermia should be monitored carefully as neonates are vulnerable for non-shivering thermogenesis and prolonged hypothermia may lead to acidosis, cardiac irritability, altered drug metabolism, delayed recovery and apneic episodes..[17]Hypothermia prevention should be taken care off. In our study, 3(3.8%) had hypoglycemia and 2 were suffered hypothermia. **Chand MB et al.** reported similar incidences of hypothermia, facial edema and dislodgement of intravenous line in 8.1% of patients.[11]The Causes for delayed recovery are prolonged action of the anaesthetic agents, inadequate reversal, hypothermia and electrolyte imbalances. In our study, 74(94.8%) had smooth and uneventful recovery, but 4(5.12%) required ventilator support. Study of **Chand MB et al.** showed smooth recovery in 32(86.4%), delayed recovery in 5(13.5%) and mechanical ventilation in 1patient(2.7%).[11]

Post-operative complications in NTD include wound infection, wound dehiscence, CSF leak, shunt infection or failure, necrotising enterocolitis, paralytic ileus and pneumonia. We noted post-operative complications in 3 cases.**Ravindra S. Giri et al.** found post-operative complications in 13 and three of them succumbed due to shock and meningitis.[12] Also, **Elmesallamy et al.** reported complications in 10patients with CSF leakage(8) and wound infection(2).[5]

CONCLUSION

In our study, children with Neural Tube Defects had several perioperative complicationsinvolvingmultiple systems. Apart from theknowledge of cerebral pathophysiology, pharmacokinetics and pharmacodynamics of anaesthetic agents the knowledge of all these perioperative complications are also needed for a meticulous anaesthetic approach for a good prognosis.

Acknowledgement: Nil

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