



ORIGINAL RESEARCH

PROFILE OF NEURAL TUBE DEFECT IN RSUD Dr. SOETOMO, 2013-2018

Profil Defek Tabung Saraf di RSUD Dr. Soetomo Tahun 2013-2018

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ABSTRACT

Background: Congenital abnormalities are structural growth abnormalities that have arisen since the intrauterine life caused by many factors, including genetics, pregnancy nutrition, infection, and social status. **Purpose:** This study aims to observe the clinical profile of Neural Tube Defect (NTD) patients in Dr. Soetomo Hospital from 2013 to 2018. **Methods:** This research was a descriptive study using secondary data from medical records from September 2013 to March 2018 to determine the profile of NTDs. The inclusion criteria for this study were patients aged 1-14 years diagnosed with NTD. Variables observed included gender, age, primary diagnosis, natal history, nutritional status, history of past therapy, referral status, comorbidities, and outputs. Data analysis was conducted in a descriptive method and presented in tables and diagrams. **Results:** This study found that out of 232 samples, 122 were female and 110 were male. Spina bifida unspecified was the most common diagnosis in 80 patients (32.78%), then encephalocele unspecified, 50 (20.49%), encephalocele anterior, 31 (12.44%), Myelomeningocele (MMC), 25 (10.24%), encephalocele unspecified, 23 (9.42%), and lipomyelocele, 20 (8.19%). There was 244 diagnosis found with 12 dual diagnoses. Works done depend on the clinical and nutritional condition of the patient. Patients with NTD tend to need more nutrition. Most NTD patients present with comorbidities, and the most common one is hydrocephalus. Many NTD patients had unknown treatment output. Most patients were still in outpatient care for further supervision. **Conclusion:** NTD incidence rate in RSUD Dr.

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Soetomo 2013-2018 is still relatively high.

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ABSTRAK

Latar Belakang: Kelainan kongenital merupakan gangguan pertumbuhan struktural yang muncul sejak kehidupan dalam rahim yang disebabkan oleh berbagai faktor meliputi genetik, gizi ibu saat hamil, infeksi dan status sosial. **Tujuan:** Penelitian ini bertujuan untuk melihat gambaran klinis pasien Neural Tube Defect (NTD) di RSUD Dr. Soetomo tahun 2013-2018. **Metode:** Penelitian ini merupakan penelitian deskriptif dengan menggunakan data sekunder dari rekam medis bulan September 2013 sampai Maret 2018 untuk mengetahui profil NTD. Kriteria inklusi penelitian ini adalah pasien berusia 1-14 tahun yang didiagnosis NTD. Variabel yang diteliti meliputi jenis kelamin, usia, diagnosis primer, riwayat kelahiran, status gizi, riwayat terapi sebelumnya, status rujukan, komorbiditas, dan keluaran. Analisis data dilakukan dengan metode deskriptif dan disajikan dalam bentuk tabel dan diagram. **Hasil:** Penelitian ini menemukan bahwa dari 232 sampel, 122 berjenis kelamin perempuan dan 110 lainnya berjenis kelamin laki-laki. Spina bifida unspecified merupakan diagnosis terbanyak, 80 pasien (32,78%), diikuti oleh encephalocele unspecified, 50 (20,49%), encephalocele anterior, 31 (12,44%), Myelomeningocele (MMC), 25 (10,24%), encephalocele unspecified, 23 (9,42%), dan lypomielocele, 20 (8,19%). Ditemukan 244 diagnosis dengan 12 diagnosis ganda. Tatalaksana bergantung pada kondisi klinis dan gizi pasien. Penderita NTD cenderung membutuhkan lebih banyak nutrisi. Sebagian besar pasien NTD datang dengan komorbiditas dan yang paling umum adalah hidrosefalus. Banyak pasien NTD memiliki luaran terapi yang tidak diketahui. Sebagian besar pasien dalam rawat jalan untuk pengawasan lebih lanjut. **Kesimpulan:** Angka kejadian NTD di RSUD Dr. Soetomo tahun 2013-2018 masih tergolong tinggi.

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INTRODUCTION

Congenital malformations are abnormalities that affect the structure and function of the existing body due to disruption of the structural growth of the fetus that develops intrauterine. Congenital malformations can be caused by single-gene defects, chromosomal abnormalities, multifactorial inheritance, environmental teratogens, and micronutrient deficiencies. Maternal infections such as rubella, maternal diseases such as diabetes mellitus, lack of iodine and folic acid, exposure to drugs and drugs including alcohol and tobacco, certain environmental chemicals, and radiation are

other factors that can cause congenital disorders (Elghanmi, Razine, Jou, & Berrada, 2020).

Every year, approximately 7.90 million babies experience congenital disorders (6% of total births worldwide). Then, severe congenital disorders occur in more than 94%, and 95% of these children die. The Southeast Asian region has the second-highest prevalence of congenital disorders globally, 9% under-five year old mortality and 12% newborn mortality. Congenital disorders are a global problem because of their impact on low and middle-income countries. Congenital malformations are considered a genetic condition, as they can cause early death and a significant

health burden. More than 90% of neonates born with congenital malformation come from low-middle income countries with limited health care costs (Umar, Salim, & Putri, 2020).

Congenital abnormalities consist of 8 types of abnormalities, including talipes, cleft lip and palate disorders, Neural Tube Defects (NTD), abdominal wall defects, anal atresia, hypospadias, epispadias, conjoined twins, and microcephaly. Trends in congenital anomalies in Europe from 1980 to 2012 were collected via EUROCAT using multiple sources of information (live births, fetal deaths (20+ weeks gestation), and pregnancy termination for fetal anomalies gestations). Among them are Microcephaly, severe Congenital Heart Disease (CHD), single ventricle, Atrioventricular Septal Defect (AVSD), Tetralogy of Fallot (TOF), Patent ductus Arteriosus (PDA), Congenital Pulmonary Cystic Adenomatous Malformation (CCAM), esophageal atresia, duodenal atresia, or stenosis-rectal atresia, and stenosis, renal dysplasia, congenital hydronephrosis (CH), leg reduction defects, club-Congenital Talipes Equinovarus (CTEV), Syndactyly, Craniosynostosis (CS) (Morris et al., 2018).

Nutrition is essential, especially in pregnancy planning or pregnancy, because a lack of nutrition can cause neural tube defects. This condition can be prevented by supplementing with folic acid every day, as much as 40 µg /day. It is proved by the decreased incidence of neural tube defects in China (Wang et al., 2016). The program to provide folic acid to expectant pregnant women is very beneficial because it is proven safe, saves money, and prevents babies' death from disabilities. If the program runs smoothly, this will help countries achieve the Sustainable Development Goals in health programs (Kancherla, Wagh, Johnson, & Oakley Jr., 2018). Good teamwork and discipline are primary in managing these congenital abnormalities. It takes a multidisciplinary approach from medical personnel and several specialists and subspecialists to allow NTD patients to provide a decent quality of life for patients (Januschek et al., 2016).

Patients with NTD need immediate therapy to improve their quality of life. In Surabaya, Indonesia, there had been no sufficient data showing the profile of NTD. This study aims to reduce the incidence of congenital abnormalities, which could reduce the morbidity, disability, and mortality in infants and children due to congenital abnormalities by observing the profile of NTD through patients referred to RSUD Dr. Soetomo.

The results of this study are expected to help raise awareness of NTD risk factors.

METHODS

This research was conducted at the Inpatient Installation of pediatrics and neurosurgery departments at RSUD Dr. Soetomo Surabaya. This research was a descriptive study using medical records to observe the profile of NTD patients. Total sampling was used. This study's inclusion criteria are pediatric patients diagnosed with NTD from 1 day to 14 years old. Variables observed included gender, age, primary diagnosis, natal history, nutritional status, history of past therapy, referral status, comorbidities, and outputs.

In this case, gender was an identity differentiated between men and women and recorded in the latest patient data and written in the medical record. Age was a unit of time to measure how long a person has lived. Comorbidities in this study were diseases other than NTD. Meanwhile, the final condition was the condition after treatment (getting better / worse. In this study, nutritional status was grouped based on the deviation of body weight for age with a z-score published by WHO, while children aged more than five years using the CDC. The WHO curve used the Z Score reference for ages 1-5 years old and CDC for ages 5-18 years old. When the LAZ value is less than -2: stunted -3: Severe Stunted, WAZ is less than -2: underweight, -3 severe underweight, WLZ and BMI for age: -2: wasted -3: severe wasted. In the WLZ and BMI for an age when the value is 1 risk of obesity, 2 overweight, 3 obese. The categorization consisted of undernutrition, normal, and malnutrition (Alemu, Ahmed, Yalew, Birhanu, & Zaitchik, 2017).

The main diagnosis basis for treatment decisions for NTD patients included Anterior Encephalocele, Posterior Encephalocele, Unspecified Encephalocele, Myelomeningocele, Lipomyelokel, Spina Bifida Unspecified. The history of therapy administration could be seen in detail based on the Medical Record's (RM) diagnostic records. The referral status was the status of the patient's arrival at Dr. Soetomo where patients came to Dr. Soetomo Hospital for the first time. They were called new patients, while patients who had undergone previous treatment but were referred to Dr. Soetomo Hospital for further treatment were referred to as referral patients. In general, referral origins are patients whose referrals are from East Java itself, such as Ngawi, Tuban, Banyuwangi, etc. Generally, the patients

are from outside East Java for the other categories, such as Lombok, Samarinda, Halmahira, etc. It was because RSUD Dr. Soetomo became a referral center from eastern Indonesia. In further therapy output distribution, the treatment for each patient is different in theory, and the management is quite different depending on the patient's condition. In the other category, most patients did not do routine control after the surgery, and the current state of the patient was unknown because not written in the medical record.

From September 2013 to March 2018, 1,085 infants were found suffering from congenital abnormalities, and 956 cases fit the inclusion criteria. Subjects with incomplete medical records were excluded from the study, leaving only 232 patients to be further observed and analyzed. Data analysis was conducted in a descriptive method and presented in tables and diagrams. This research has received a certificate of ethical clearance by the Health Research Committee of RSUD dr. Soetomo Surabaya with ethical number 1582 / KEPK / X / 2019.

RESULTS

This study is a descriptive retrospective study using medical records of patients with NTD in Dr. Soetomo from 2013 to 2018. Based on the study results, 244 samples met the inclusion and exclusion criteria, with 12 duplicate diagnoses found, leaving 232 samples for further analysis. Table 1 shows six types of diagnoses found in this study. The most common diagnosis in this study was Spina Bifida in 80 patients (32.78%). Of the total 232 patients in the study, 12 patients (4.92%) were diagnosed with multiple diagnoses, so the entire diagnosis found was 244 (Table 1).

Table 1
Diagnosis

Characteristics	n	%
Diagnosis per person		
Single Diagnosis	232	95.08
Multiple Diagnosis	12	4.92
Diagnosis Diseases		
Anterior Encephalocele	46	18.85
Posterior Encephalocele	23	9.42
Unspecified Encephalocele	50	20.49
Myelomeningocele	25	10.24
Lipomyelocele	20	8.19
Unspecified Spina Bifida	80	32.78
Total	244	100.00

Table 2 explained the combination of multiple diagnoses found in 12 patients. The most common combination was anterior encephalocele with unspecified spina bifida and unspecified encephalocele with Myelomeningocele (MMC), both 3 patients (25%). Posterior and non-specific encephalocele with spina bifida ranks second-most, 2 patients (16.66%) of both, followed by encephalocele unspecified with lipomyelocele and posterior encephalocele, both accounting for 1 patient (5.88%) (Table 2)

Table 3 showed that NTD patients in Dr. Soetomo were dominated by children in the age group of 0-5 months, 100 patients (43.10%), followed by children aged 6 months or older, 49 patients (21.12%), 6-11 months, 29 patients (12.5%), 12-17 months old, 16 patients (6.89%), and 18-23 months, 14 patients (6.03%). NTD occurred in less than 3 percent in children aged 24-29 months, 6 patients (2.58%), 36-41 months, 5 patients (2.15%), 30-35 months 4 patients (1.72%), 48-53 months, 3 patients (1.29%). and least occurred in children aged 42-47 months, with only 2 patients having NTD (0.81%). More females 122 (52.58%), suffered from NTD than males, 110 (47.41%), with a ratio of 1: 1.10 (Table 3).

Table 2
Combination of Diagnosis

Characteristics	n	%
Anterior Encephalocele + Unspecified Spina Bifida	3	25.00
Unspecified Encephalocele + MMC	3	25.00
Posterior Encephalocele + Unspecified Spina bifida	2	16.66
Unspecified Encephalocele + Unspecified Spina Bifida	2	16.66
Posterior Encephalocele + MMC	1	8.33
Unspecified Encephalocele + Lipomyelokel	1	8.33
Total	12	100.00

NTD patients were dominated by referrals from hospitals in Surabaya, with 62 patients (26.72%), hospitals in Madura with 57 patients (24.56%), self-aware 49 patients (21.12%), hospitals outside java with 47 patients (20.25%), hospitals in Probolinggo, 9 (3.87%) and from hospitals in Sidoarjo 8 patients (3.44%). NTD patients were dominated by 128 (55.17%) undernutrition patients, followed by 71 patients (30.60%) with normal nutritional status and 33 patients (14.22%) who suffered from malnutrition.

Most patients in this study were first diagnosed in RSUD Dr. Soetomo and came at various ages depending on the type and severity of the neural tube diseases. Table 3 showed that in 80 spina bifida unspecified cases, mostly came at age 0-5 months (31 patients) or over 60 months (25 patients); 50 encephalocele unspecified cases mostly came in the age of 0-5 months (23 patients); and 46 anterior encephalocele cases mostly came in over 60 months old (11 patients). Then, there was 25 myelomeningocele cases mostly went in the age of 0-5 months (12 patients); a total of 23 posterior encephalocele case mostly went in the period of 0-5 months (16 patients), and a total of 20 lipomyelocele cases mostly went in the age of 0-5 months (8 patients). Most NTD patients tend to be undernutrition (Table 3).

There were 76 patients who came with a previous comorbid disease (30.40%), 156 others with no comorbidities (60.40%), and 18 with multiple comorbidities. The most common comorbidities were hydrocephalus with 41 patients (43.61%), then lacrimal system disorder, 14 patients (14.89%), Congenital Talipes Equinovarus (CTEV), 11 patients (11.70%), epilepsy, 9 patients (9.57%), Atrial Septal Defect (ASD), 8 patients (8.51%), bladder dysfunction, 7 patients (7.44%) and Ventricular Septal Defect (VSD), 4 patients (4.25%). Twenty-nine Spina bifida unspecified cases have the highest comorbidities, and 14 patients suffered from hydrocephalus. The least comorbid found in posterior encephalocele (Table 4).

There were 6 treatments and workups found. Some patients received multiple treatments and workups, making up a total of works done by 383 actions. Clean wound surgery in 125 patients (33.63%) were the most work performed in all diagnosis except in meningomyelocele (Table 5) and followed by regular outpatient done in 74 patients (19.72%), cele excision in 66 patients (17.23%), myelocyte encephalocele excision in 47 patients (12.27%), gross wound surgery in 47 patients (12.27%). The least work performed was Ventriculoperitoneal (VP) shunt in 24 patients (6.26%).

Medical care was given by outpatient control to 62 patients (25.40%), 6 patients (2.45%) returned home, 10 patients died (4.09%), 3 patients (1.22%) were cared for in inpatient care, and patients with other final conditions were accounted for 163 patients (66.81%). Unspecified spina bifida mostly has unknown output (62 patients); it

can instead be an unknown or lost to follow up; while lipomyelocele cases were also mostly unknown (13 patients) (Table 6).

DISCUSSION

The youngest patient treated at Dr. Soetomo Hospital was 1 day old, while the oldest was 14 years old. A similar research was conducted at the Pakistan National Institute of Child Health affiliated with the Department of Neurosurgery, Pediatric Neurosurgery Subdivision. It contained the youngest age was 4 days old, and the oldest was 1.30 years old (Rehman, Farooq, & Bukhari, 2018). This study found that patients who came with a diagnosis of NTD were mostly in 0-5 months. NTD is a common defect in humans. Around 300,000 neonates are born with NTD globally each year. Over the past few decades, there have been significant reductions in NTD incidents around the world, but they are still high in developing countries (Zaganjor et al., 2016).

The incidence of NTD in children less than a year old because of NTD at birth can be distinguished by inspection, but still have to use CT scans and MRI to diagnose. The authors acquired data that there are patients who survive and do routine control for up to 14 years. It is almost the same as research from various sources, such as PubMed by Matos namely about 76% of deaths occur in the first day of life, about 71% survive until the age of 1 year, and 67% until the age of 20 (Cruz & De Jesus, 2021). The incidence of NTD can be affected by factors such as lack of folic acid intake, family risk factors, maternal diabetes, maternal hyperthermia, smoking, alcohol consumption, premature birth, CMV infection, etc. (Dawood, 2019).

In the current study, it was found that most NTD patients were female, with a male-to-female ratio of 1: 1,12. This result is similar to the study in Jinnah Postgraduate Medical Center, Karachi, Pakistan, with 17 males and 33 females (Rehman, Farooq, & Bukhari, 2018). There were 400 cases of frontoethmoidal encephalocele in the other cases, of which 212 men and 188 women. This research was conducted at Soetomo General Hospital, Surabaya, and the Charity Foundation Program from 2008 to 2015 was reviewed (Arifin, Suryaningtyas, & Bajamal, 2018).

Table 3
Sociodemographic Characteristics Distribution

Characteristics	Encephalocele		Unspecified		Myelocele		Total	
	Anterior	Posterior	Encephalocele	Spina Bifida	Meningo-	Lipo-	n	%
Age (months)								
0-5	10	16	23	31	12	8	100	43.10
6-11	4	2	10	8	1	4	29	12.50
12-17	5	1	3	6	4	0	16	6.89
18-23	7	1	2	2	1	1	14	6.03
24-29	2	0	2	1	0	1	6	2.58
30-35	4	0	0	1	0	0	4	1.72
36-41	2	1	1	1	0	0	5	2.15
42-47	1	0	0	0	1	0	2	0.86
48-53	0	0	1	2	0	0	3	1.29
54-59	0	0	0	3	0	0	4	1.72
> 60	11	2	8	25	6	6	49	21.12
Gender								
Male	22	8	23	39	17	13	110	47.41
Female	21	13	23	39	7	7	122	52.58
Nutritional Status								
Malnutrition	5	4	10	5	2	9	128	55.17
Undernutrition	25	10	22	16	8	54	71	30.60
Normal	16	9	18	4	10	17	33	14.22
Referral origins								
Surabaya	17	6	21	45	12	10	111	47.84
Madura	19	5	13	15	4	1	57	24.56
Sidoarjo	1	2	2	1	0	2	8	3.44
Probolinggo	0	0	3	2	2	2	9	3.87
Other	6	8	7	15	6	5	47	20.25
Total	43	21	46	78	24	20	232	100.00

Table 4
Comorbidities Distribution

Comorbidities	Encephalocele		Unspecified		Myelocele		Total		
	Anterior	Posterior	Encephalocele	Spina Bifida	Meningo-	Lipo-	n	%	
Hydrocephalus		6	3	8	14	5	5	41	43.61
Lacrimal System Disorder		8	0	2	4	0	0	14	14.89
CTEV		0	0	1	6	2	2	11	11.70
Epilepsy		2	1	0	5	1	0	9	9.57
ASD		1	0	2	3	1	1	8	8.51
Bladder Dysfunction		0	1	1	1	3	1	7	7.44
VSD		1	0	2	0	1	0	4	4.25
Total		18	5	16	33	13	9	94	100.00

Table 5
Treatments and Workups Distribution

Treatments & Workups	Encephalocele		Unspecified		Myelocele		Total	
	Anterior	Posterior	Encephalocele	Spina Bifida	Meningo-	Lipo-	n	%
Clean Wound Surgery	21	15	28	41	10	10	125	32.63
Gross Wound Surgery	5	4	12	12	13	1	47	12.27
Cele Excision	13	9	13	16	12	3	66	17.23
Myelocele	10	5	9	13	4	6	47	12.27
Encephalocele Excision								
VP Shunt	4	4	1	7	7	1	24	6.26
Regular outpatient	16	5	13	30	4	6	74	19.32
Total	46	23	50	80	25	20	383	100.00

Table 6
Therapy Output Distribution

Output	Encephalocele		Unspecified		Myelocele		Total	
	Anterior	Posterior	Encephalocele	Spina Bifida	Meningo-	Lipo-	n	%
Outpatient Control	15	5	13	16	8	5	62	25.40
Inpatient Care	0	0	2	0	1	0	3	1.22
Died	4	0	0	1	0	1	10	4.09
Returned Home	1	4	2	1	1	1	6	2.45
Other	26	14	33	62	15	13	163	66.81
Total	46	23	50	80	25	20	232	100.00

The main NTD diagnosis in this study included anterior encephalocele, posterior encephalocele, unspecified encephalocele, unspecified spina bifida, myelomeningocele, and lipomyelocele. The most found cases were unspecified spina bifida, and the least was lipomyelocele. The multiple diagnosis combinations found were anterior encephalocele with unspecified spina bifida and unspecified encephalocele with MMC. Spina bifida was a congenital disorder in which the spinal cord did not develop properly due to incomplete closure of the neural tube at about 28 days of gestation. From 2005 to 2010, Githuku had 1296 births diagnosed with NTD, with 1184 cases of spina bifida and 8 cases of encephalocele. Of 1,184 spina bifida, 18 cases were found with hydrocephalus, and 12 cases were found with CTEV (Githuku et al., 2014).

Encephalocele is a congenital abnormality characterized by herniation of the brain due to brain deficiencies in the bones of the skull. Congenital disorders can occur because a closure failure of neuroporus anterior is not closed within 26-28 days of pregnancy (Arifin, Suryaningtyas, & Bajamal, 2018). In a research conducted in the

Philippines from 2010-to 2013, it was found 30 cases of frontoethmoidal encephalocele (Marshall, Setty, Hnatiuk, & Pieper, 2017).

Research in China between 2014-2019 found 11 cases of spinal meningocele. Six are Caudal Types, 2 Para-neural Types, and 3 Neural Types. The meningocele in types I and II was tied after no nerve involvement was found. For Type III, the herniated sac and involved nerve roots are tied when the nerve root is indicated as malfunctioning on monitoring (Cheng et al., 2020).

In MMC, myeloma and meninges were exposed to abnormal vertebral arches development in early pregnancy. Myelomeningocele (MMC) is the most common type of NTD from spinal defects. It occurred with extrusion of the spinal cord and/or meninges into the sac containing cerebrospinal fluid (CSF) through a defect in the vertebral arch (Peranteau & Adzick, 2016). Usually, patients with myelomeningocele would experience motor, sensory, and neurological deficits depending on the location of the defect. Research conducted by North at the British Columbia the Children's Hospital (BCH) Spinal Cord Clinic between 1971 and 2016 found 309 patients diagnosed with myelomeningocele. It is an increase from previous studies (North, Cheong, Steinbok, & Radic, 2018).

Lipomyelocele is an occluded NTD that contains fat and myeloma herniation, similar to lipoma. At the same time, lipomas itself are masses of fat tissue and fibrous in the vertebral column through spina bifida defects. Lipomyelocele is very rare compared to MMC (Ogawa et al., 2007). Only 3 cases were found from a total of 23 NTD patients.

The management of NTD patients depends on the clinical conditions and comorbidities of the patient. Each diagnosis had at least two different treatments and workups done in this study. There were 44 NTD patients with hydrocephalus comorbidity. Moreover, hydrocephalus was found the most in encephalocele with a total case of 12, followed by 10 spina bifida cases. Of all NTDs with hydrocephalus, 23 of them were performed VP shunt to reduce the incidence of CSS leakage, protect the brain from hydrocephalus, shorten hospital stay, and raise the cost-efficiency. Still, the risk of infection in the VP shunt is rather high, and surgery in patients with lowered immunity is not yet optimal. Yorulmaz and Konak found that 102 of 140 patients suffering from hydrocephalus with spina bifida were performed VP shunt (Yorulmaz & Konak, 2019). Cele excision is commonly performed in all NTD cases. Wohon found that main management in hydrocephalus cases with encephalocele was an excision of the cele and VP shunt that requires a long surgical time and precise preparation (Wohon, Harijono, & Saleh, 2012).

Developing countries dominate the largest number of NTD. It occurs due to a lack of education before pregnancy, which causes congenital disorders (Lee, Byun, Nguyen, Schlosser, & Gudis, 2020). However, in the current study, the origin of NTD patients was still dominated by referrals from the hospitals in Surabaya, with many from hospitals in Madura, Sidoarjo, Probolinggo, and outside East Java. In other therapy, output distribution is after the patient underwent treatment that is not routinely controlled or not written in the medical record. It is due to the role of Dr. Soetomo hospital as a referral hospital for Eastern Indonesia. 2018 Ministry of Health Survey found that there are still many morbidity cases in Indonesia, ranked in the top 10. The incidence in this study is similar to East Java incidence (Ilmi, Kalanjati, & Suryaningtyas, 2019).

Most NTD patients come with low nutritional status. In Turkey, the research found five encephalocele patients aged 0 months, three

patients with undernutrition and two patients with malnutrition (Ugras, Kavak, Alpay, Karabekir, & Bicer, 2016). From 2003 to 2016, 86 patients aged 4 to 16 years in Malaysia found that the lowest body weight was around 1.80 kg, and the heaviest was 4.60 kg (Sahmat et al., 2017). Although, a case report from Yokohama reported that neonatal encephalocele patients could also occur in neonates weighed 3.80 kg with a normal weight-for-age and weight-to-height scale (Pahlevi, Heryani, & Zuhro, 2019).

Most NTD patients experience comorbid diseases other than Arnold Chiari, one of which is hydrocephalus. Arnold Chiari is a neurosurgical disorder due to malformations caused by abnormalities through the foramen magnum, reaching the spinal canal, so the defect builds up and results in a decrease in the brain stem, abnormalities of the 4th ventricle and lateral ventricles through the foramen magnum. Hydrocephalus is a build-up of excess cerebrospinal fluid in the brain due to order, flow, or absorption abnormalities. It generally occurs in 80% of individuals with myelomeningocele or encephalocele. Patients with comorbidities have a higher mortality rate. MMC is the most severe subtype of spina bifida. It is known to lead to neurogenic bladder and more severe paralysis or sensory loss that can cause death if not treated immediately. Hydrocephalus have neural networks' presence is significant with neurological deficits that can happen due to thromboses primary therapy, and others due to ventriculoperitoneal shunt complications during abdominal surgery. Also, premature babies increase the risk of ASD and VSD. Patients with multiple diagnoses generally increase a higher mortality rate (Dicianno, Sherman, Roehmer, & Zigler, 2018), and 140 of 186 patients (Yorulmaz & Konak, 2019). In encephalocele, 4 of 40 patients (Rehman, Farooq, & Bukhari, 2018) and 10 of 27 encephalocele patients suffer from hydrocephalus (Yucetas & Uçler, 2017).

Congenital abnormalities, particularly neural tube defects with other comorbidities, require expensive treatment and a multidisciplinary approach involving neonatologists/pediatricians, radiologists, physiotherapists, obstetricians, neurosurgeons, and psychologists to optimize patient outcomes. However, with the costs that must be paid, the patients are forcibly discharged and usually have a terrible social stigma for the condition (Upeh, Ijasan, & Eze, 2019).

Kadia, Aroke, Tianyi, Bechem, & Dimala (2017) reported a case of a 22-year-old 36 weeks pregnant woman with non-optimal antenatal care, who performed CS during the labor expulsion phase. It results in a normal first and second twin with spina bifida, hydrocephalus, and bilateral CTEV. The first neonate was in normal conditions in other multiple labors, but the second one had an infected open spina bifida with hydrocephalus and CTEV comorbidities. All reported died due to the comorbid.

Neurosurgeons have found a way to increase the survival rate in NTD patients. More recently, the MOMs (Management of Myelomeningocele Study) trial reported randomized evidence regarding the efficacy of prenatal repair, opening the possibility for improved outcomes and a potential reduction for CSF shunting, but at the risk of an increase in prenatal complications and preterm birth. The exceptional cost and limited availability of intrauterine MMC closure made it unlikely to exert a significant impact. Because the neurosurgical intervention cannot restore function loss due to proper neural tube closure failure, all neurosurgical treatment of open NTDs remains fundamentally palliative. Therefore, primary prevention remains the most important and effective tool (Estevez-Ordóñez et al., 2018).

Research Limitations

Data retrieval was only based on medical records as secondary data. It makes researchers unable to review the condition and treatment of the patients directly. Another limitation besides patients rarely do routine control is incomplete data, such as the patient's current condition after treatment and whether he survives. Hopefully, we can find out the survival rate with this data, which can be used as additional information about neurosurgery.

CONCLUSION

The incidence of NTD in Dr. Soetomo during 2013 to 2018 is still quite high. Unspecified spina bifida was the most common diagnosis in 80 patients (32.78%), then encephalocele unspecified, 50 (20.49%), encephalocele anterior, 31 (12.44%), Myelomeningocele (MMC), 25 (10.24%), encephalocele unspecified, 23 (9.42%), and lypomielocele, 20 (8.19%). There was 244 diagnosis found with 12 dual diagnoses. Works done depend on the clinical and nutritional condition of the patient. Patients with NTD tend to need more nutrition. Most NTD patients present

with comorbidities, and the most common one is hydrocephalus. Many NTD patients had unknown treatment output. Most patients were still in outpatient care for further supervision.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest in this study.

AUTHOR CONTRIBUTION

All authors participated actively in the research. MAP, PIG, WS: conceptualization, methodology, and supervision. MNA.: data curation, draft preparation, reviewing, and editing.

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