

Frequency of Hydrocephalus in Patients Presenting with Spinal Dysraphism

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ABSTRACT

Objective: The objective of this study is to determine the frequency of hydrocephalus in patients presenting with spinal dysraphism.

Material and Methods: This cross sectional descriptive study was conducted at Department of Neurosurgery, Postgraduate Medical Institute Lady Reading Hospital, Peshawar, Pakistan. A total of 119 patients of spinal dysraphism were included through convenience (non-probability) sampling, in a period of six months (from June, 2011 to December 2011). All patients with spinal dysraphism with either sex and age less than 2 years included, while patients with post spinal dysraphism surgery who developed hydrocephalus, posterior fossa lesion causing obstructive hydrocephalus and Patients with post tuberculous meningitis with hydrocephalus were excluded. Hydrocephalus was observed in these patients. The data was analyzed using the statistical program SPSS version 17.

Results: Out of total number of 119 patients, majority was males with 66 in number (55.5%), while female were 53 (44.5%). The age of patients ranged from 10 days to 23 months with overall mean age 5.47 ± 5.439 months. Majority of patients, 82 (68.9%), were in the age range of 01-06 months. Most of the patients, i.e., 56 (47.1%) were harboring spinal dysraphism at lumbar region. Hydrocephalus was found in quite large number of cases, 79 cases (66.4%), in patients of spinal dysraphism.

Conclusion: Spinal dysraphism is slightly more common in males with frequency of 55.5%. Majority of the patients of spinal dysraphism (68.9%) were presented in the age range of 1 – 6 months. Spinal dysraphism is more common (47.1%) at lumbar region. Hydrocephalus was found in 79 cases (66.4%) of spinal dysraphism.

Key words: Hydrocephalus, Spinal Dysraphism, Spina Bifida, Lumbar region.

Abbreviations: MMC = Myelomeningocele.

INTRODUCTION

Spinal dysraphism (spina bifida) also known as neural tube defects are congenital malformations of the spine and spinal cord secondary to abnormal neural tube closure that occur between the third and fourth weeks of gestation. The term spinal dysraphism includes the overall group of defects derived from the maldevelopment of the ectodermal, mesodermal, and neuroectodermal tissues, and its sequelae may affect brain, bones, extremities, and bowel and bladder functions.¹

The cause of spina bifida is multifactorial and includes genetic predisposition, nutritional deficiencies, particularly folate and zinc, use of anti-epileptic drugs like carbamazepine or valproic acid, diabetes mellitus (type – 1), pre-pregnancy obesity and possibly other non medical factors such as agricultural pesticides, radiation, hyperthermia and use of tobacco or drugs.² Different forms of spina bifida can be distinguished, varying from mild to severe. The most common form is myelomeningocele.³ Patients with Spina

Bifida also tend to be less adaptable, easily distracted, less attentive and persistent, and less predictable.⁴

The prevalence of spina bifida has declined in developed countries of the world owing to both prenatal folate supplementation and to pregnancy termination following prenatal diagnosis. In United States before 1980 prevalence of spina bifida was 1-2/1000 live births, but more recently prevalence has declined to 0.44 per 1000 live births. But unfortunately, in third world countries prevalence is much higher, and acceptable prevalence data are not available, nor has the issue been addressed with the goal of eradication, or reduction of incidence.²

The spinal dysraphism is associated with other central nervous system abnormalities like hydrocephalus, chiari II malformation, and spinal cord dysplasia.^{3,5} Hydrocephalus is a clinical condition characterized by increased amount of cerebrospinal fluid in the brain. It results in dilatation of ventricles, increase intracranial pressure, brain atrophy, neurological impairment and even death.^{6,7} The cognitive outcome in spina bifida has frequently been attributed to the severity of the hydrocephalus.³

Hydrocephalus was diagnosed in 88% of the patients of spinal dysraphism and all were treated with the insertion of a ventriculoperitoneal (VP) shunt. In 79.7% the shunt was placed simultaneously before repair of myelomeningocele at one sitting and in 20.2% the VP shunt was placed after repair.² Various studies have shown different prevalence rate of hydrocephalus in patients presented with spinal dysraphism, i.e. 50%,¹ 75%,¹ 80%,⁸ 88%,² 90%.⁴ The potential for prevention of the spina bifida like neural tube defects in the third world countries is still far from being fulfilled and there is great need to increase awareness among the general public regarding etiological factors of this disastrous anomaly and its prevention by appropriate prenatal care.²

The current study is designed to determine the frequency of hydrocephalus among patients presenting with spinal dysraphism. The rationale behind doing this study is to generate local statistics of hydrocephalus among spinal dysraphism patients as no local data is available on this aspect. The results of this study will be shared with other neurosurgeons to make them aware about the local magnitude of the problem and on the basis of results of this study if the frequency of hydrocephalus is found in significant number of patients then we will be able to recommend not only early screening of all patients with spinal dysraphism for hydrocephalus but also early referral to tertiary care.

MATERIAL AND METHODS

This cross sectional descriptive study was conducted at Department of Neurosurgery, PGMI Lady Reading Hospital, Peshawar – Pakistan. The sample size was 119, keeping 50% proportion of hydrocephalus among spinal dysraphism, 95% confidence interval and 9% margin of error using WHO sample size calculation and consecutive non probability sampling technique was opted. The study duration was 6 months (June 2011 to December 2011).

Inclusion Criteria

All patients with spinal dysraphism with either sex and age less than 2 years were included.

Exclusion Criteria

Patients with post spinal dysraphism surgery who developed hydrocephalus, patients with posterior fossa lesion causing obstructive hydrocephalus diagnosed on CT and those with post tuberculous meningitis with hydrocephalus as diagnosed on medical records and history were excluded.

After permission from hospital ethical committee, patients with spinal dysraphism presenting to Neurosurgery Department of Lady Reading Hospital through outpatient and emergency department were enrolled. Those who fulfill inclusion criteria were included in the study. Informed consent was taken from all patients. These patients were further assessed through detailed history, including personal particulars, name, age, sex, address, symptoms and through detailed clinical examination. Diagnosis of spinal dysraphism were made on the basis of posterior midline swelling anywhere from cervical up to sacral region, since birth and presence of any one of these: lower limb weakness of less than MRC Grade 5, hypertrichosis or hemanomatous discoloration.

From all patients' detailed history, detailed neurological and systemic examination and Computerized tomography of brain were done to diagnose hydrocephalus. All CT scan were reported by a single expert radiologist, the fellow of CPSP. All the observation and examination were done by me and all data were recorded in a predesigned Performa. Strictly exclusion criteria were followed to control confounders and bias in the study results.

The data was analyzed using the statistical program SPSS version 17. Descriptive statistics like mean \pm standard deviation were used for age. Frequency / percentage was calculated for categorical variables

like, sex, site of lesion and hydrocephalus. Hydrocephalus was stratified among the age, gender and site of lesion to see the effect modifiers. All results were presented in the form of tables, charts and graphs for different variables.

RESULTS

Sex Incidence

Out of total number of 119 patients, majority was males with 66 in number (55.5%), while female were 53 (44.5%) with male to female ratio of 1.25:1.

Age Range

The age of patients ranged from 10 days to 23 months. In this study the overall mean age was 5.47 ± 5.439 months. Majority of patients, 82 (68.9%) were in the age range of 1 – 6 months, followed by patients in age group of < 1 month (Table 1).

Site Involved

In this study majority of the patients 56 (47.1%) were harboring spinal dysraphism at lumbar area, followed by lumbo-sacral, dorso-lumbar, dorsal, cervical and finally cervico-dorsal region (Figure 1).

Table 1: Age – Wise Distribution of Patients (n = 119).

	Frequency	Percent	Valid Percent	Cumulative Percent
< 1 month	15	12.6	12.6	12.6
1 – 6 months	82	68.9	68.9	81.5
7 – 12 months	12	10.1	10.1	91.6
> 1 year	10	8.4	8.4	100.0
Total	119	100.0	100.0	

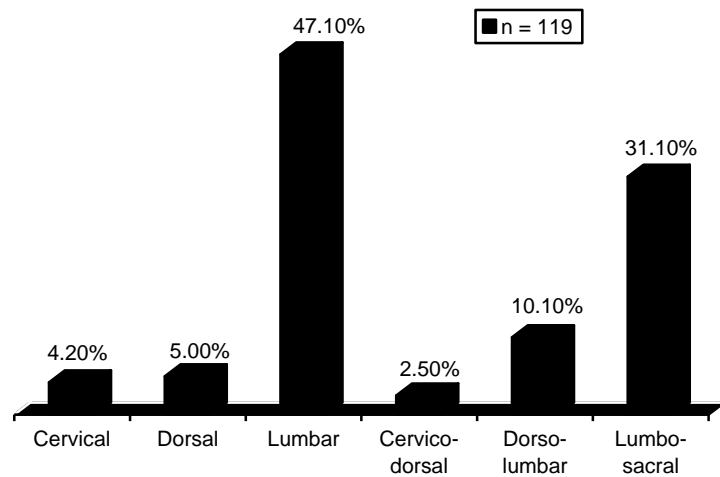


Figure 1: Site of Lesion (n = 119).

Incidence of Hydrocephalus

Hydrocephalus was found in quite large number of cases, i.e., 79 cases (66.4%) in patients of spinal dysraphism, while remaining 40 (33.6%) patients were not having hydrocephalus.

Geographical Data

Geographical status of the patients showed that most of the patients 30 (25.2%) belonged to the capital of province, i.e., Peshawar, followed by Charsadda, Mardan and Swabi, 17 cases to Kohat, Karak, Bannu, D.I Khan, 11 cases each of boner, dir and agencies, 10 cases belonged to Chamkani, Nowshera, 9 cases to

Table 2: Address – Wise Distribution of Patients (n = 119).

	Frequency	Percent	Valid Percent	Cumulative Percent
Peshawar	30	25.2	25.2	25.2
Chamkani, Nowshera	10	8.4	8.4	33.6
Shangla, Swat, Malakand	9	7.6	7.6	41.2
Kohat, Karak, Bannu, DI Khan	17	14.3	14.3	55.5
Charsadda, Mardan, Swabi	26	21.8	21.8	77.3
Boner, Dir	11	9.2	9.2	86.6
Agencies	11	9.2	9.2	95.8
Punjab, Sindh	2	1.7	1.7	97.5
Afghanistan	3	2.5	2.5	100.0
Total	119	100.0	100.0	

Table 3: Stratification of Age (Presence of Hydrocephalus in Patients with Spinal Dysraphism) (n = 79).

			Age Range				Total
			< 1 Month	1 – 6 Months	7 – 12 Months	> 1 Year	
CT Findings	Hydrocephalus = Yes	Count	10	52	10	7	79
		% within CT Findings	12.7%	65.8%	12.7%	8.9%	100.0%
	Hydrocephalus = No	Count	5	30	2	3	40
		% within CT Findings	12.5%	75.0%	5.0%	7.5%	100.0%
Total		Count	15	82	12	10	119
		% within CT Findings	12.6%	68.9%	10.1%	8.4%	100.0%

Table 4: Stratification of Site of Lesion (Presence of Hydrocephalus in Patients with Spinal Dysraphism) (n = 79).

			Site of Lesion						Total
			Cervical	Dorsal	Lumbar	Cervico-dorsal	Dorso-lumbar	Lumbo-sacral	
CT Findings	Hydrocephalus = Yes	Count	3	4	32	2	10	28	79
		% within CT Findings	3.8%	5.1%	40.5%	2.5%	12.7%	35.4%	100.0%
	Hydrocephalus = No	Count	2	2	24	1	2	9	40
		% within CT Findings	5.0%	5.0%	60.0%	2.5%	5.0%	22.5%	100.0%
Total		Count	5	6	56	3	12	37	119
		% within CT Findings	4.2%	5.0%	47.1%	2.5%	10.1%	31.1%	100.0%

swat, Shangla, Mardan, 3 cases belonged to across the border, Afghanistan and only 2 patients belonged to other provinces of the country, one each from Punjab and Sindh (Table 2).

In majority of cases i.e., 62 (52.1%), neurology was impaired, while neurology was intact in 57 (47.9%) cases.

Management

In the current study most of the patients, 75 cases (63%), were offered surgery, of these 75 cases surgery for hydrocephalus i.e., ventriculo-peritoneal shunt was done in 50 cases (42%), while myelomeningocele

surgery, excision and repair was done in 24 cases (20.2), simultaneous myelomeningocele (MMC) repair and VP shunt done in 1 (0.8) case and rest of the patients 44 (37%) were treated conservatively.

Relationship of Hydrocephalus and Age

Stratification of hydrocephalus among age was done and it showed that majority of the patients, 52 (65.8%), with hydrocephalus was between 1 – 6 months age range, followed by 10 patients in the age range of < 1 month and 7 – 12 months each and finally rest of the patients of hydrocephalus, 7, was in the age range of > 1 year (Table 3).

Hydrocephalus and Site of Dysraphism

When hydrocephalus was stratified among the site of lesion of spinal dysraphism, so it was found that most of the patients of hydrocephalus having spinal dysraphism at lumbar region, followed by lumbo-sacral, dorso-lumbar, dorsal, cervical and finally hydrocephalus was present in patients of spinal dysraphism at cervico-dorsal region (Table 4).

DISCUSSION

Spinal dysraphism is one of the most common congenital abnormalities encountered by all neurosurgeons and they need to focus not only on the view under microscope but also on a view outside microscopic field of vision. We should focus on the disorders that affect large numbers of children.⁹

The prevalence of myelomeningocele has declined in developed countries, like USA and most of the European countries of the world because of the fact that both prenatal folate supplementation and the pregnancy termination following prenatal diagnosis have played a very important role. In United States before 1980 prevalence of myelomeningocele was 1-2/1000 live births, but more recently it has declined to 0.44 per 1000 live births.¹⁰⁻¹²

While, in third world countries the prevalence of spinal dysraphism is much higher owing to the lack of above mentioned factors, mainly due to the poverty and low literacy rate.

Furthermore, in our society the issue of abortion is controversial. There are two schools of thoughts regarding this, first thought is of those who say that the abortion is illegal or forbidden and they blame religion for this, which is not a true scenario. The other group says that abortion is the fundamental right of a couple if they had found some antenatal defects in the fetus, but the ratio of this group is quite low. Another reason for not aborting the child is that the facility of antenatal neurosurgical consultation is not available in the rural areas of the city and we got main bulk of the patients from those areas.

Above mentioned reasons are the some facts that the prevalence of spinal dysraphism is not declining in our part of the world as compared to developed countries.

Spinal dysraphism occurrence is equal or somewhat more common in females (female, 1.0 – 1.7/ male, 1.0), depending on the populations studied.¹ In the current study the number of male patients was slightly high; with 66 in number (55.5%), while female

were 53 (44.5%). When compare with a national study done by Hashim et al, 2 out of 415 cases there were 205 (49.3%) males and 210 (50.7%) girls. So, the data is somewhat compatible with the literature.

Spinal dysraphism can present at any age, like the open type presents or manifested right after the birth, because of the apparent swelling at the back, but on the other hand the occult or closed type can present late, at any age, because of the tethering of the spinal cord.¹³

The research showed that most of the patients of occult spinal dysraphism present in the first 6 months of age and this is the best time to operate upon them.¹³ In the current study the age of patients ranged from 10 days to 23 months and when we compare the presenting age of our patients so, majority of patients 82 (68.9%) were in the age range of 1 – 6 months, which is compatible with the literature.¹³

Another study done on patients of tethered cord syndrome showed the presenting age of patients was in the range of 7 – 15 years,¹⁴ but in our study the age range is 10 days – 23 months. This is because of the fact that we included both types of spinal dysraphism and secondly we included only those patients whose age was less than 2 years.

Geographical distribution of the patients showed that only 30 patients (25.2%) belonged to Peshawar, while the major bulk i.e., 89 patients (74.78%) belonged to other parts of the KPK province and other provinces and Afghanistan. This data emphasize the fact that there should be neurosurgical tertiary care facilities in the major cities of the province, to decrease the workload on a single tertiary care hospital.

In an international study the frequency of spinal dysraphism at different sites was as; cervical = 0 – 5%, dorsal = 5 – 10%, thoraco-dorsal = 20 – 30%, lumbar = 20 – 30%, lumbo-sacral = 30 – 50% and sacral 3 – 15%.⁸ In the current study the major bulk {56 patients (47.1%)} was formed by the patients having spinal dysraphism at lumbar area, followed by lumbo-sacral area 37 patients (31.1%), which is comparable with the literature.¹⁵

Various studies have shown different prevalence rate of hydrocephalus in patients presented with spinal dysraphism, i.e. 50%,¹ 75%,¹ 80%,⁸ 88%,² 90%.⁴

We found hydrocephalus in large number of cases of spinal dysraphism, i.e., 79 cases (66.4%). This frequency of hydrocephalus is comparable with national and international literature, as literature showed frequency of hydrocephalus in the range of 50 – 90%.^{1-2, 4, 8}

In almost two third, 62 (52.1%), of our cases neurology was impaired, either in the form of lower limb weakness and/or sphincteric dysfunction. In a series by a national author² showed 155 (37.3%) patients having myelodysplasia, were paraplegic and incontinent. When we look at the international literature, the frequency of impaired neurology ranges from 44-79% (44%,¹⁶ 55%,¹⁷ 57%,¹⁸ 78%,¹⁹ 79%²⁰).

In the current study most of the patients, 75 cases (63%), were offered surgery, of these 75 cases surgery for hydrocephalus i.e., either ventriculo-peritoneal shunt, external ventricular drain or therapeutic CSF tap was done in 50 cases (42%), while myelomeningocele surgery, excision and repair was done in 24 cases (20.2), simultaneous MMC repair and VP shunt done in 1 (0.8) case and rest of the patients 44 (37%) were treated conservatively. The reason being not operating upon 37% of the patients was multiple, like either patient was having infection (treated conservatively and later definite surgery was planned when infection was clear) or there is no benefit upon operating on the patient, because of the lorber's criteria.²¹

We counsel all those parents whose children were paraplegic and explained them that the spinal dysraphism is the lifelong commitment and there is also the development of the risk of hydrocephalus and other neurological deficits. And usually most of the parents, they submit to all these consequences. They are guided and directed through regular physiotherapy and rehabilitation centers.

Stratification of hydrocephalus among age was done and it showed that majority of the patients, 62 (78.5%), with hydrocephalus was in the first 6 months of their life. Literature also showed that in patients of spina bifida, hydrocephalus is apparent in the first few weeks of life.^{1,11,15,22}

In this study, hydrocephalus, when stratified among gender, it was found to be somewhat equal among both genders, i.e., 39 (49.4%) males and 40 (50.6%) females were having hydrocephalus among the total patients. From a study done by WHO showed that there is male predominance in the patients of hydrocephalus alone, but when hydrocephalus is associated with the spina bifida, the male: female falls to unity, means there is equal male to female predominance.²³

When hydrocephalus was stratified among the site of lesion of spinal dysraphism, so it was found that most of the patients, 32 (40.5%), of hydrocephalus having spinal dysraphism at lumbar region, followed by lumbo-sacral region having 28 (35.4%) patients, dorso-lumbar region with 10 (12.7%) patients, dorsal

region with 4 (5.1%) patients, cervical region with 3 (3.8%) patients and finally hydrocephalus was present in patients of spinal dysraphism at cervico-dorsal region with 2 (2.5%) patients. So, it is evident that occurrence of hydrocephalus is more in the upper lesions, i.e., lumbar and above. In our study it is almost 2/3rd (64.6%) of the patients having hydrocephalus associated with spinal dysraphism at or above lumbar region.

This data is compatible with Netto JMB et al,¹ which proved that in his patients about 90% of the patients have hydrocephalus and the spinal level of spinal dysraphism was at or above lumbar level.

In perspective of the study results, I would like to make few recommendations. Firstly it is obvious that about 2/3rd, 79 (66.4%), of the patients of spinal dysraphism has hydrocephalus, which is a significant number of patients, so it is recommended to do not only early screening of all patients with spinal dysraphism for hydrocephalus but also early referral to tertiary care hospital.

Second important thing is that spinal dysraphism is a serious and devastating anomaly having other associated congenital disorders and it is lifelong commitment with overall not fruitful prognosis. One should opt the different measures to prevent spinal dysraphism, as famous saying "Prevention is better than cure". Primary prevention requires the prevention of neural tube defects occurring in the embryo in the first place. Since the 1960s, it has been recognized that women with an affected pregnancy had significantly lower red blood cells folate level than those with unaffected pregnancies.

The Medical Research Council Vitamin Study of 1991,²⁴ proved that women who had had a previous pregnancy affected with a neural tube defect were randomized to receive folic acid (4 mg daily) or placebo, with or without other multivitamin supplements, demonstrated that the rate of affected subsequent pregnancy was significantly reduced in the folic acid group.

This fact is now recommended that in order to prevent a recurrence of a neural tube defect in subsequent pregnancies, 5 mg daily of folic acid should be supplemented to the diet prior to conception and, therefore, in order to prevent a first occurrence of a neural tube defect, all women should be advised to take 400 mg of folic acid daily prior to conception, as well as increasing their dietary intake of foods rich in folic acid, continued until the termination of first trimester of pregnancy. Foods high in folate includes green vegetables,

yeast, beef extract and breakfast cereals fortified with folic acid.²⁵ Secondary prevention of further affected pregnancies requires screening techniques, including AFP sampling and ultrasound, with selective termination of future affected pregnancies.²⁵

CONCLUSIONS

From the results of this study it is concluded that:

- Spinal dysraphism is slightly more common in males with frequency of 55.5%.
- Majority of the patients of spinal dysraphism (68.9%) were presented in the age range of 1-6 months.
- Spinal dysraphism is more common (47.1%) at lumbar region.
- Hydrocephalus was found in 79 cases (66.4%) of spinal dysraphism.
- Frequency of hydrocephalus in patients of spinal dysraphism was somewhat equal in both genders, i.e., 49.4% in males and 50.6% in females.
- Spinal dysraphism associated with hydrocephalus was more common (40.5%) at lumbar region.

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