

**Original Article****Lipomyelomeningocele: Epidemiological Studies in a Pediatric Neurosurgery Department of Bangladesh**Rahman MN<sup>1</sup>, Mukherjee SK<sup>2</sup>, Arman DM<sup>3</sup>, Ekramullah SM<sup>4</sup>, Rashid MMH<sup>5</sup>, Ansari A<sup>6</sup>, Ziauddin M<sup>7</sup>, Mahbub AA<sup>8</sup>, U Chowdhury<sup>9</sup>, Shykot ZH<sup>10</sup>, Rahman MM<sup>11</sup>, Muktadir MR<sup>12</sup>**Conflict of Interest:** There is no conflict of interest relevant to this paper to disclose.**Funding Agency:** Was not funded by any institute or any group.**Contribution of Author:** Principal Investigator- Dr. Md. Nafaur Rahman, Manuscript Preparation- Dr. Sudipta Kumer Mukherjee, Dr. D. M. Arman, Prof. Sk. Md. Ekramullah

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**Copyright:** ©2021 bang.BJNS published by BSNS. This published by BJNS. This article is published under the creative commons CC-BY-NC license. This license permits use distribution (<http://creativecommons.org/licenses/by-nc/4.0/>) reproduction in any medium, provided the original work is properly cited and is not used for commercial purposes.**Received:** 08 October, 2021  
**Accepted:** 15 October, 2021**Abstract****Background:** Spinal dysraphism, one of the most common birth defects, has been estimated to occur annually in more than 320,000 infants worldwide. Lipomyelomeningocele (LipoMMC) is a closed neural tube defect in which the surface lipomatous tissue is attached to the spinal cord. LipoMMC results from premature disjunction of the epithelial ectoderm from the neural ectoderm at a gestational age of 18–28 postovulatory days. Both genetic and environmental factors are suggested to have important roles in the occurrence of neural tube defects. In addition, variables such as vitamin B 12 deficiency, maternal hyperinsulinemia, maternal dietary glycaemic intake, maternal obesity and use of antiepileptic medications during pregnancy have been suggested as risk factors for spinal dysraphism.**Objective:** The aims and objectives of this study was to see the epidemiology of Lipomyelomeningocele in our population. We shall also compare our result with other previous study result.**Materials and method:** It is a retrospective study. Cases were obtained from a NINS Pediatrics neurosurgery electronic registry ([emrbd.com](http://emrbd.com)). The study was carried out at Department of Paediatrics Neurosurgery, NINS&H, and Dhaka from November 2017 to till date. Subjects was included from a NINS Pediatrics neurosurgery electronic registry. Structured questionnaire was used to collect the necessary information. Risks and benefits of this study were explained to the patient and patient's legal guardian/parents in an easily understandable local language.**Conclusion:**

In this study the majority of cases were female. Environmental factors also appear to be associated with lipomyelomeningocele risk factor. Additional studies needed to establish the environmental risk factor associated with LipoMMC.

**Key words:** Lipomyelomeningocele, Neural tube defects, Pediatric, Epidemiological, Bangladesh.**Abbreviations:** IBM: International Business Machines

Lipo MMC: Lipomyelomeningocele

NTD: Neural tube defects

NINS&amp;H: National Institute of Neuro Sciences &amp; Hospital

SES: Socio-economic status

SPSS: Statistical Package for the Social Sciences

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**Introduction:**

Spinal dysraphism, one of the most common birth defects, has been estimated to occur annually in more than 320,000 infants worldwide<sup>1</sup>. It is classified as open or closed dysraphism<sup>2</sup>. Open spinal dysraphisms include meningocele, myelomeningocele, myeloschisis, encephalocele, and anencephaly. All involve exposure of nervous tissue and/or meninges to the external environment<sup>3</sup>. Closed spinal dysraphisms are known as lesions with covered neural tissue<sup>4</sup>. Those are frequently accompanied by cutaneous stigmata such as dimples, hemangiomas, soft tissue masses, lipomas, and hairy nevi<sup>5,6</sup>.

Lipomyelomeningocele is a type of congenital occult spinal dysraphism or spina bifida consisting of the presence of lipomatous (fatty) tissue attached to the dorsal spinal cord, which protrudes through a spinal defect along with the meninges or spinal cord to form a posterior mass below the skin, usually in the lumbosacral region<sup>7</sup>. Lipomyelomeningocele (LipoMMC) is a closed neural tube defect in which the surface lipomatous tissue is attached to the spinal cord. LipoMMC can present as only a skin lesion without any neurological deficits or with neurological disorders like lower-extremity weakness and/or deformity and bladder dysfunction secondary to a tethered cord<sup>8</sup>.

LipoMMC results from premature disjunction of the epithelial ectoderm from the neural ectoderm at a gestational age of 18–28 postovulatory days<sup>9</sup>. The embryology of the defect is thought to involve an anomaly in primary neurulation, in which the neural ectoderm separates from the cutaneous ectoderm. As a result, periaxial mesoderm comes in contact with the unfused ventral neural ectoderm. The mesoderm then differentiates into fatty tissue, thus preventing the neural canal and the posterior aspect of the spine from fusing<sup>10-11</sup>.

The higher frequency of neural tube defects (NTD) malformations secondary to abnormal neural tube closure that occur between the third and fourth weeks of gestational age among children born to women of lower socio-economic status (SES) has been well documented in the epidemiological literature<sup>12</sup>.

Lipomyelomeningocele comprises 8–25% of all cases of spina bifida<sup>13-15</sup>. The lipomyelomeningocele rate has been estimated to be 2.5 per 10,000 births by clinical series studies<sup>11, 16</sup>, although one population based study reported a rate of 1.6 per 10,000 births<sup>17</sup>. The defect is found more frequently among females<sup>13-17</sup>.

Maternal periconceptional use of folic acid has been determined to reduce the risk of NTDs among their offspring<sup>18</sup>. Conversely, some studies have reported no significant reduction in the rate of LipoMMC following folic acid fortification<sup>7, 17, 19</sup>. However, a study in Nova Scotia, Canada, also reported no reduction in lipomyelomeningocele rates subsequent to folic acid fortification in that country<sup>17</sup>. Esmaeili et al., 2013 observations suggest that the pathogenesis and risk factors of LipoMMC may differ from those of other spinal dysraphisms<sup>8</sup>.

Forrester & Merz, 2004 observation also may indicate that the pathogenesis of lipomyelomeningocele may differ from myelomeningocele. It also reported higher lipomyelomeningocele rates in infants born to mothers in younger and older age groups<sup>7</sup>.

Both genetic and environmental factors are suggested to have important roles in the occurrence of neural tube defects. Not much is known about the correlation of genetics and the incidence of LipoMMC, although it seems that there must be a genetic contribution to the occurrence of this defect as with some other kinds of neural tube defects in siblings of LipoMMC-affected patients<sup>8</sup>.

Sarris, Tomei, Carmel, & Gandhi, 2012 also reported that neural tube defects have a complex origin in which both environmental and genetic factors play a role<sup>3</sup>. Maternal obesity has been associated with an increased risk of neural tube defects, suggesting that the embryonic environment for development may be metabolically different for obese women<sup>20-21</sup>. Familial forms of lipomyelomeningocele are rare, with only 2 previous reports<sup>5</sup>.

In addition, variables such as vitamin B 12 deficiency, maternal hyperinsulinemia and use of antiepileptic medications during pregnancy have been suggested as risk factors for spinal dysraphism<sup>22</sup>. Maternal dietary glycemic intake has been suggested as risk factors for spinal dysraphism in some studies<sup>23-25</sup>.

Here we study about epidemiological study of LipoMMC in Pediatric Neurosurgery department of Bangladesh.

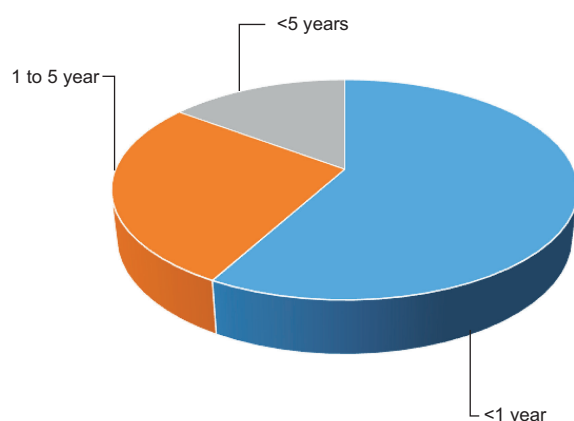
**Materials and Methods:**

The aims and objectives of this study was to find out epidemiology of lipomyelomeningocele in our population. We also compared our result with other previous study result. The study was carried out at

Department of Paediatrics Neurosurgery, NINS&H, and Dhaka from November 2017 to till date. This study was a retrospective study. Data collection sheet was used to collect the necessary information. Subjects was included from a NINS&H Pediatrics neurosurgery electronic registry. Structured questionnaire was used to collect the necessary information. Risks and benefits of this study were explained to the patient and patient's legal guardian/parents in an easily understandable local language. Informed written consent was taken from each guardian before data collection. Statistical analysis was done by using SPSS software version 24.0. (IBM). Statistical significance will be set at p-value <0.05 at 95% level of confidence interval.

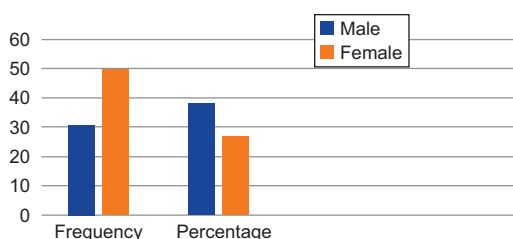
### Results:

Figure 1 shows distribution of the study patients according to age. Here most of the patients were within <1 year age group 47 (58.0%). Second large were within 1-5 years age group 22 (27.2%).



**Fig.-1:** Distribution of study subjects according to age (n=81)

Figure 2 shows distribution of the study subjects according to gender. About 31 (38.3%) patients were males and 50 (61.7%) patients were females.



**Fig.-2:** Distribution of study subjects according to gender (n=81)

Table I shows distribution of the study subjects according to geographical distribution. Here most of the patient hailing from Dhaka division 38 (46.9%). About 12 (14.8%) patient hailing from Chittagong division.

**Table-I**

*Distribution of subjects according to geographical area (n=81)*

Geographical area	Frequency (n)	Percentage
Rangpur division	5	6.2
Rajshahi division	7	8.6
Mymensing division	3	3.7
Sylhet division	0	0
Dhaka division	38	46.9
Khulna division	7	8.6
Barisal division	9	11.1
Chittagong division	12	14.8

### Discussion:

In a population-based study of LipoMMC, Esmaeili et al., 2013 reported an increased prevalence among females; they found that 68.6% of cases were female.

In our study we also found most of the patients are females 61.7%. Other most the studies also show an increased prevalence among females.

Both genetic and environmental factors are suggested to have important roles in the occurrence of neural tube defects. In general, neural tube defects have a complex origin in which both environmental and genetic factors play a role<sup>3</sup>. But not much is known about the correlation of genetics and the incidence of LMMC<sup>8</sup>.

In our studies we also found most of the patient hailing from most densely populated and industrial division of Bangladesh. About 46.9% patient hailing from Dhaka division and about 14.8% patients hailing from Chittagong division.

Dhaka division is the main central industrial zone in Bangladesh. Along with the great growth in industry in the region, there have also been increased and sometimes critical levels of toxic pollution to the environment<sup>26</sup>. Chittagong is the second largest city in Bangladesh and has a population of approximately 4.5 million people. The city hosts a large seaport and extensive industrial activities in and around the city and subsequent emissions of pollutants to the environment<sup>27</sup>.

So there may be an environmental factor for developing of most LipoMMC in these two division.

### Conclusion:

In this study the majority of cases were female. Environmental factors also appear to be associated with lipomyelomeningocele risk factor. Additional studies needed to establish the environmental risk factor associated with LipoMMC.

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