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Omphalocele and Gastroschisis: Comparison of Outcome in A Resource Limited Tertiary Centre

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Background. Neonates with gastroschisis are expected to have better prognosis than omphalocele as the latter is commonly associated with other congenital anomalies. But in our centre, we experience the opposite scenario regarding outcome.

The aim of this study was to compare the outcome of these two conditions and to some extent to identify the factors influencing the consequences.

Methods. It was a prospective observational study done at Dhaka Shishu (Children) Hospital from June 2017 to November 2017. All neonates admitted with omphalocele and gastroschisis during the study period were included. Data were collected in a structured questionnaire.

Results. Total number of cases were 38 (24 omphalocele & 14 gastroschisis). None of the patients were antenatally diagnosed. Gender, mean birth weight, mean gestational age, maternal age and mode of delivery demonstrated inconsiderable influence on the outcome. Out of 24 patients with omphalocele, in 20 patients, it was associated with other anomalies, and the other 4 patients died before evaluation. Mortality rate was significantly higher in gastroschisis (86%) than with omphalocele (42%).

Conclusion. Inadequate perinatal management contributes to the poorer prognosis of gastroschisis in our centre. Antenatal diagnosis, planned delivery and appropriate management in immediate post natal period can improve the result of these conditions.

Key words: Neonates, Omphalocele, Gastroschisis, outcome.

Омфалоцеле та гастрошизис: порівняння результатів лікування у ресурсо-обмеженному третинному центрі

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Огляд: Вважається, що новонароджені з гастрошизісом мають кращий прогноз, ніж з омфалоцеле, оскільки останнє захворювання здебільшого супроводжується іншими вродженими аномаліями розвитку. Але в нашому центрі спостерігається протилежний сценарій відносно результатів лікування. Метою даного дослідження було порівняння результатів лікування цих двох захворювань і певною мірою визначити чинники, що впливають на результати лікування.

Методи: Нами проведено обсерваційне дослідження в дитячій лікарні Дака Шішу з червня по листопад 2017 р. У дослідження були включені всі новонароджені, які поступили з омфалоцеле та гастрошизисом протягом періоду дослідження. Дані були зібрані в стандартизованій анкеті.

Результати: Загальна кількість випадків становила 38 (24 дитини з омфалоцеле та 14 - з гастрошизисом). У жодного пацієнта діагноз не встановлено антенатально. Такі показники як гендерне співвідношення, середня вага при народженні, середній гестаційний вік, вік матері та спосіб пологів не мали значного впливу на результат. З 24 дітей з омфалоцеле у 20 пацієнтів захворювання супроводжувалось іншими аномаліями розвитку, а інші 4 пацієнта померли до проведення обстеження. Рівень смертності був значно вищий (86%) при гастрошизисі, ніж при омфалоцеле (42%).

Висновок: Невідповідне перинатальне ведення сприяє гіршому прогнозуванню при гастрошизисі в нашому центрі. Антенатальна діагностика, плановані пологи та відповідне лікування в безпосередньо постнатальному періоді можуть покращити результати цих захворювань.

Ключові слова: новонароджені, омфалоцеле, гастрошизис, результати.

Омфалоцеле и гастрошизис: сравнение результатов лечения в ресурсо-ограниченном третичном центре

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Обзор: Считается, что новорожденные с гастрошизисом имеют лучший прогноз, чем с омфалоцеле, так как последнее заболевание чаще всего сопровождается другими врожденными аномалиями развития. Но в нашем центре наблюдается противоположный сценарий относительно результатов лечения.

Целью данного исследования было сравнить результаты лечения этих двух заболеваний, а также определить факторы, влияющие на результаты лечения.

Методы. Нами проведено обсервационное исследование в детской больнице Дака Шишу с июня по ноябрь 2017 г. В исследование были включены все новорожденные, поступившие с омфалоцеле и гастрошизисом в течение периода исследования. Данные были собраны в стандартизованной анкете.

Результаты. Общее количество случаев составило 38 (24 ребенка с омфалоцеле и 14 - с гастрошизисом). Ни у одного пациента диагноз не установлен антенатально. Такие показатели как гендерное соотношение, средний вес при рождении, средний гестационный возраст, возраст матери и способ родов не имели значительного влияния на результат. Из 24 детей с омфалоцеле у 20 пациентов заболевание сопровождалось другими аномалиями развития, а другие 4 пациента умерли до проведения обследования. Уровень смертности был значительно выше (86%) при гастрошизисе, чем при омфалоцеле (42%).

Вывод. Ненадлежащее перинатальное ведение способствует худшему прогнозированию при гастрошизисе в нашем центре. Антенатальная диагностика, планирование родов и соответствующее лечение непосредственно в постнатальном периоде могут улучшить результаты этих заболеваний.

Ключевые слова: новорожденные, омфалоцеле, гастрошизис, результаты.

Introduction

Omphalocele and gastroschisis are the commonest among the anterior abdominal wall defects in neonates. These two anomalies exhibit different pathogenesis. In omphalocele, viscera herniate through umbilical ring with a membrane covering; while in gastroschisis viscera herniate through a gap, usually to the right of the umbilical cord and not covered by membrane [1]. Gastroschisis usually presents as an isolated defect, though 10%–20% babies with this pathology have intestinal anomalies like atresia, volvulus, gangrene etc. These conditions are recognized as complicated gastroschisis and associated with dreadful prognosis [1-3].Children with omphalocele are generally associated with other congenital abnormalities, particularly chromosomal and cardiac anomalies. These accompanying anomalies bear importance in determining the outcome of a patient with omphalocele [1,4]. The developed countries have been achieving greater success in managing these grave conditions through the improvement of perinatal care. However, babies with these congenital anomalies, still remain as notable causes of morbidity and mortality in developing countries [5]. Reports from developed countries showed higher morbidity and mortality among babies with omphalocele as this condition is often associated with other abnormalities [4,6,7]. As an isolated anomaly, neonates with gastroschisis are supposed to have a better consequence, but previous study from our centre and also from other developing countries, reflected worse outcome in babies with gastroschisis due to poor perinatal management [8].

Table 1
Demographic data

Variables	Omphalocele (n=24)	Gastroschisis (n=14)	p
Birth weight (kg)	2.62±58	2.17±27	.06
Gestational age (weeks)	36.16±1.65	35.78±1.36	.26
Maternal age (years)	22.00±2.90	20.71±3.79	.70
Gender (M/F)	M-12, F-12	M-9, F-5	.50
Mean antenatal ultrasound	1.83±.64	1.92±92	.70
Antenatal diagnosis	Nil	Nil	

In this study we made an effort to point out the factors responsible for the poorer outcome of the patients with gastroschisis and omphalocele in Bangladesh.

Materials and methods

It was a prospective observational study held in Dhaka Shishu (Children) Hospital from June 2017 to November 2017. All neonates admitted with omphalocele and gastroschisis during the study period were included. Demographic and clinical data were collected in a structured questionnaire. SPSS version 22 was used for statistical analysis. Continuous data were presented as mean ± SD and analysed by student t-test. Categorical data were presented as frequency and analysed by Chi square test. p value <.05 considered significant for both test.

Results

Total 38 patients were admitted with omphalocele and gastroschisis during these six months. Among them

Table 2
Associated anomalies

Omphalocele minor (n=11)	Omphalocele major (n=9)	Ruptured omphalocele (n=4)	Gastroschisis(n=14)
Meckels band-2 Intestinal atresia- 3 Cardiac anomaly- 11	Cardiac anomaly- 9	Not evaluated	Ileal atresia- 1

Table 3
Treatment & mortality

	Omphalocele minor (n=11)	Omphalocele major (n=9)	Ruptured omphalocele (n=4)	Gastroschisis (n=14)	P
Treatment	Primary repair -10 Escharosant- 1	Escharosant – 9	Reposition – 3 Silo – 1	Silo- 5 Repair – 7 No -2	
Mortality	4	2	4	12	.01

24 were with omphalocele and the rest of them with gastroschisis. Every mother had at least 1 ultrasound scan during pregnancy but fetal abnormality was unnoticed. Male babies were 55.26% (12 with omphalocele & 9 with gastroschisis), 44.74% were female (12 with omphalocele & 5 with gastroschisis). Mean birth weight was 2.62±58 kg in omphalocele and 2.17±27 kg in gastroschisis. Mean gestational age of babies with omphalocele & gastroschisis were 36.16±1.65 & 35.78±1.36 weeks respectively. Mean maternal age was 22±2.9 & 20±3.8 years in omphalocele and gastroschisis, respectively. Associated anomalies were present in 83.33% (20 out of 24) of neonates with omphalocele, the others died before evaluation. Babies with gastroschisis were not evaluated for associated anomalies. Mortality in gastroschisis was 85.71% (12 out of 14) and in omphalocele was 41.67% (10 out of 24).

Discussion

Dhaka Shishu (Children) Hospital is renowned as the largest specialized paediatric hospital in Bangladesh. We receive patients with omphalocele and gastroschisis referred from all over the country.

Increasing incidence of anterior abdominal wall defect has been reported around the world [4,5], however, in our country their prevalence has not been surveyed yet. An integrated protocol has become a crying need to provide quality care to these newborn babies.

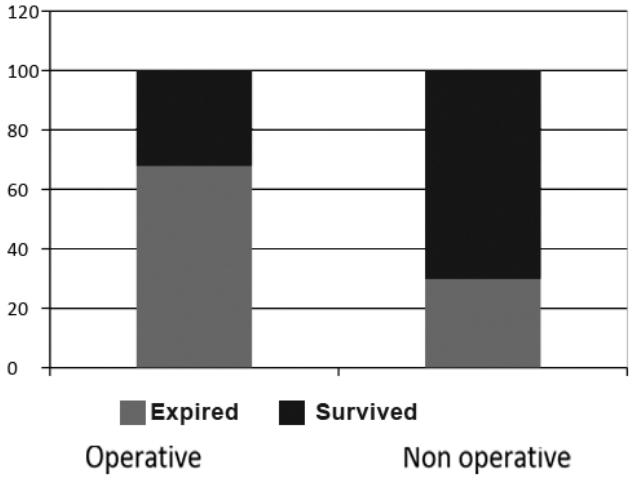

Fig. Operation and survival

Unfortunately, not a single baby was diagnosed during antenatal checkup, though every mother had at least one ultrasound scan during pregnancy. This result reflects inadequate exposure & experience of radiologists regarding these pathologies. Similar finding was reported in Nigeria by Abdur-Rahman L O et al. [5], while in developed countries, almost 100% cases are detected prenatally that contributes to the excellent outcome by optimizing the time & place of delivery and postnatal management [4,9,10].

Demographic data showed insignificant statistical difference between omphalocele & gastroschisis; however the mean birth weight in gastroschisis was lower. A larger sample size would have altered this finding to a statistically distinct one. Watanabe S et al found this difference noteworthy [7] in his study. Many authors identified low maternal age as a risk factor for gastroschisis [4,7,11,12]. However, our study identified the maternal age irrelevant, which precisely agrees with Abdur-Rahman LO et al. [5]

All of the babies with omphalocele, we could evaluate, were with cardiac anomalies (predominantly ASD & PDA). This is a common finding [1,4,7]. But in this study, the babies with omphalocele minor were noted to have associated intestinal anomalies. This phenomenon is not designated as a frequently observed one. Four babies with omphalocele major died before evaluation for cardiac anomalies. All of them had ruptured sac. Only one baby with gastroschisis had intestinal atresia. We could not investigate the babies with gastroschisis for associated anomalies as they died before evaluation.

Most of the neonates with omphalocele minor underwent primary repair as the content was small and there was associated intestinal anomalies. Nine cases of omphalocele major had non-operative management with escharosant initially as non-operative management of omphalocele major is encouraged in resource limited centres to avoid post operative complications arising from raised intra-abdominal pressure [5]. Sac was ruptured in one case among them and the patient could not survive after repair.

Almost all of the babies with gastroschisis presented to us more than 12 hours after delivery with exposed

oedematous viscera. The babies were hypovolemic, hypothermic, even in shock. Two babies died during resuscitation. After reposition of the gut, these babies developed respiratory failure and ultimately died as we do not have facilities for elective ventilation. Problem encountered in patients with ruptured omphalocele were not any different.

Previous study from same centre identified delayed presentation and inappropriate postnatal management along with limited resource are responsible for poor outcome of gastroschisis. Despite the fact that initial survival rate was significantly higher in patients with omphalocele, data regarding long-term survival and quality of life are yet to be investigated.

Moreover, deaths occurring at home and on the way to health facility due to these conditions are hardly recorded, which creates a hindrance to reach the goal of Every Newborn Action Plan (ENAP), launched by United Nations Children's Fund (UNICEF), recommending counting of every birth as well as death is essential to reduce preventable neonatal death [13]. Much better data and identifying the factors contributing neonatal death are crucial to attain the ENAP target of less than 12 neonatal deaths per 1000 by 2030. Without encountering the prevalence of gastroschisis and omphalocele, it is highly unlikely to raise awareness and promote targeted actions which are indispensable for avoiding delayed management.

Conclusion

Incompetent antenatal investigations and lack of immediate postnatal care ensuing worse initial prognosis in gastroschisis compared to omphalocele, although it is often complicated by other congenital anomalies. Multidisciplinary integrated protocol is required for better outcome of gastroschisis and omphalocele.

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