A Retrospective Review of Gastroschisis at The University Hospital of the West Indies: Factors affecting outcome

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ABSTRACT

Objective: To identify the antenatal, perinatal and postnatal factors associated with adverse outcomes in infants diagnosed with gastroschisis at The University Hospital of the West Indies (UHWI) over a ten-year period.

Methods: A retrospective review of all cases of pregnancies complicated by gastroschisis over a ten-year period at the UHWI was conducted. The primary objective of this study is to determine the antenatal factors that affect adverse perinatal outcomes of neonates with gastroschisis. The composite adverse neonatal outcome comprised of death, preoperative and postoperative wound infection, sepsis, ventilator support, parenteral nutrition, bowel resection and the presence of hernia one-year post repair. Univariate and multivariate analysis were utilized to assess factors associated with adverse outcomes.

Results: There were 14 infants with gastroschisis at The University of the West Indies from 2006–2016 included in the study. Among the cohort, six (42%) were born at the institution (inborn) while nine (64%) were transferred after birth (outborn) to UHWI for further management. Within the cohort 11 (79%) had one or more adverse outcome. Four infants died during their admission, giving a mortality rate of 29%. The period diagnosis (antenatal *vs* postnatal), birthweight, antenatal ultrasound surveillance and place of birth (Inborn *vs* outborn) were variables identified as predictors of adverse outcomes.

Conclusion: Antenatal diagnosis and birth in a tertiary centre, are factors that may be implemented to improve the outcomes of infants affected by gastroschisis.

Keywords: Abdominal wall defects, gastroschisis, University of the West Indies

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INTRODUCTION

Gastroschisis is the most common congenital anterior abdominal wall defect and in the majority of cases is an isolated defect (1). Various segments of bowel and other abdominal organs, such as the liver and spleen may also be herniated through the defect (2). There may be associated complications including intrauterine growth restriction, prematurity, fetal demise, bowel torsion, atresia, necrosis and short bowel Syndrome, parenteral nutrition and assisted ventilation. The incidence of gastroschisis has increased in many parts of the world over the last 2–3 decades (3, 4) and the prevalence of is now approximately one in 4000 live births (5). Although there is an apparent increase in the incidence of gastroschisis worldwide, marked improvement in infant mortality has been noted with early prenatal diagnosis with fetal surveillance and improved neonatal care in general (2–4, 6). Morbidity is strongly influenced by the presence of bowel pathology at birth [atresia, necrosis, perforation, or volvulus] (7).

Mortality ranges between 8% and 10% in developed countries. In third world or developing countries, mortality rates of 43–79% have been experienced (8, 9). In a recent retrospective review done at the Bustamante Hospital for Children (the largest paediatric referring intuition in Jamaica), the mortality rate was quoted to be 79% (9). Factors that have been identified to be associated with increased mortality in underdeveloped countries include the absence of prenatal diagnosis, prematurity, low birthweight, delivery outside a tertiary center, delayed surgical intervention (8–10), as well as the lack of preformed silos, parenteral nutrition and intensive care facilities (11).

With widespread availability of maternal screening and ultrasound prenatal diagnosis enables counseling, expertise involvement of interdisciplinary teams, early intervention and proper planning for delivery, all which have been proven to offer better perinatal outcomes (12). Ideally,

delivery should occur in a tertiary care facility with appropriate resources for caring for these neonates as the weight of evidence suggests inborn newborns have better outcomes than those who require transfer after birth for treatment (11). The prenatal detection rate of gastroschisis has been estimated at between 76–90% but in regions with a good routine ultrasound programme detection rates approach 100% (13).

This study seeks to examine the mortality and morbidity of infants diagnosed with gastroschisis as well as to determine if there is difference in outcome between those infants born at the institution compared to those born outside. This study is the first review of the outcomes at at The University Hospital of the West Indies (UHWI) and it seeks to identify factors deemed responsible for adverse perinatal outcomes in our neonatal population. This may enable UHWI to identify areas deficient in antenatal care, obstetric and neonatal practices in an effort to increase survival rates and improve outcomes in neonates diagnosed with gastroschisis.

SUBJECTS AND METHODS

A retrospective review of all cases of gastroschisis from January 1, 2006 to December 31, 2016 at the UHWI was carried out. The cases were identified from records from the Labor Ward, Special Care Nursery and an ultrasound database (Viewpoint ™ General Electric). Maternal and neonatal variables were obtained from the medical records of the cases identified. Cases with other fetal abnormalities detected in the antenatal period and incomplete data were excluded from this study.

Maternal data collected included demographic characteristics, medical history, antenatal care and ultrasound data. Perinatal data and adverse outcomes (including death, sepsis, wound infection, bowel complications) were also recorded.

The means and standard deviations of continuous variables were reported. Maternal, perinatal and neonatal variables were then statistically analysed to determine if there are any associations with adverse outcomes. Univariate analysis was performed using Chi-square tests and where more than two cells were projected; Fisher's exact test was used to assess the relationship between variables. A *p*-value of < 0.05 was considered statistically significant. Multivariate (logistic regression) analysis was also used to assess factors associated with adverse outcomes.

RESULTS

There were 14 infants with gastroschisis managed at the UWHI from 2006 to 2016 included in the study: six (42%) were born at the institution (inborn) while nine (64%) were transferred after birth (outborn) to the U.H.WI. for management.

The mean maternal age was 23.2 ± 5.6 years and nine (64%) of the women were primigravid. All women denied smoking, drug or alcohol use during pregnancy and had no comorbid illnesses. Of the 14 cases only five (36%) had an antenatal diagnosis of gastroschisis while the remainder nine (64%) were diagnosed in the postnatal period. Most of the women 12/14 (86%) had attended 12 or more antenatal visits. In the group diagnosed in the postnatal period, 8/9 (89%) had no obstetric ultrasound during the pregnancy. These cases were all outborn. Only 1/14 (7.1%) had less than three ultrasounds and 5/14 (35.7%) having more than three ultrasounds.

The bowel changes noted on ultrasound were not stated in the charts reviewed. Of those with an antenatal diagnosis, 4/5(80%) had antenatal consultation and counseling by a paediatric surgeon prior to delivery. Only 2/14 (14%) were documented as having intra-uterine growth restriction and one (7%) was found to have polyhdramnios. There were no cases of oligohydramnios identified. The mean gestational age of delivery was 37.3 ± 2 weeks. The majority, 10/14 (71%) was *via* spontaneous vaginal delivery and 4/14 (29%) *via* Cesarean delivery.

Of the neonates seven (50%) were male and seven (50%) female. The mean birthweight was 2.4 kg ± 430 grams; seven (50%) were term and six (43%) were preterm. Only one (7%) had Apgar scores less than seven at five minutes. Paediatric surgery consultation was given within 12 hours after birth in 11 (78%) of the cases: 10/14 (71%) of neonates had an abdominal defect ≥ 4.0 cm. Only one infant was admitted to the intensive care unit. Primary fascial closure was achieved in seven (50%) of cases and staged delayed closure in the remaining seven (50%).

Of the 14 neonates in the cohort, 11/14 (79%) had one or more adverse outcome. The need for parenteral nutrition was the most common adverse outcome (11/14 infants [79%]) followed by sepsis (9/14, 64%). Four infants died (all outborn), giving a mortality rate of 29%. Three of those (75%) neonates died of sepsis making it the most common cause of death. The other was due to respiratory failure of a preterm neonate. Six cases (43%) had a hernia one-year after closure of the defect. Other complications included preoperative wound infection 5/14 (34%), postoperative wound infection, 5/14 (34%) and bowel resection 4/14(29%). 4/14 (29%) infants required ventilator support. The average time to commence enteral feeds was 17.4 days.

On Bivariate analysis, preoperative and postoperative wound infections as well as the need for ventilator support were influenced by antenatal and postnatal factors. The cases diagnosed in the postnatal period had a greater proportion of preoperative infection (71%) than those diagnosed

in the antenatal period (0%, $p = 0.03$). Those neonates with a birthweight ≥ 2.5 kg had less need for ventilator support (0%) than those of low birthweight < 2.5kg (57 %, $p = 0.018$). Those who did not have antenatal ultrasounds had more preoperative wound infection (71 %) than those who had greater than three ultrasounds (0%, $p = 0.03$).

The inborn infants overall had better outcomes than the outborn infants. Inborn infants had no preoperative wound infection (0%) compared to the outborn group (71%, $p = 0.028$). Additionally, the majority of outborn infants also developed a wound infection post repair compared to inborn (55.6% *vs* 0%, $p = 0.038$). Outborn infants had a much higher mortality rate 44% compared to inborn (0%) although not statistically significant ($p = 0.08$).

DISCUSSION

In the previous study in Jamaica (9), the high mortality (79%) was attributed to prematurity, complicated gastroschisis, lack of parenteral nutrition and inadequate transfer protocols (9). They also concluded that improving antenatal care and establishing transfer protocols might improve the outcome in Jamaica. In our study the lower mortality (29%) may due to management in tertiary care that offers a multidisciplinary approach to the management allowing adequate preparation for delivery.

Numerous studies have attempted to identify reliable prenatal predictors to minimize poor neonatal outcomes (14, 15). Perinatal predictors previously investigated include intrauterine growth restriction, oligohydramnios, polyhydramnios, small for gestational age, five minute Apgar score and the presence of other fetal anomalies (16–19). In our study, none of these variables were

predictive of adverse neonatal outcomes. However, those neonates with a birthweight ≥ 2.5 kg had less need for ventilator support (0%) than those of low birthweight < 2.5 kg (57 %, $p = 0.018$). This is not surprising as the smaller size infant often has inadequate lung maturity. More frequent prenatal ultrasounds were found to be associated with improved outcomes. Those with multiple ultrasounds likely had earlier antenatal diagnosis allowing for preparation for delivery and surgery.

Delivery of patients diagnosed with gastroschisis, diagnosed in the antenatal period, should occur in a facility with appropriate resources for caring for these neonates as much evidence suggests inborn newborns have better outcomes than those who require transfer after birth for treatment (11, 20). Our study highlighted that adverse outcomes and increased mortality rates are greater amongst outborn infants when compared to inborn although not proven to be statistically significant. Inborn infants had no preoperative wound infection compared to outborn (0% *vs* 71%, $p = 0.028$). This may also be the contributing factor to the fact that the majority of outborn infants also developed a wound infection post repair of their defect compared to inborn (55.6% *vs* 0%, $p = 0.038$). This may have been attributed to conditions in the place of birth; issues with transfer possibly leading to more hypoxic injury. Although not statistically significant, outborn infants nine (64%) were more likely to require ventilator support than those born at UHWI (44% *vs* 0%, p-value = 0.078). Therefore it is not surprising that outborn infants had a much higher mortality rate 44% compared to inborn (0%) although this was not proven to be statistically significant ($p = 0.08$).

The mortality rate was 0% for all inborn patients. These neonates benefitted from immediate paediatric surgical care resulting in earlier closure of their defect, less need for ventilator support, bowel complications, parenteral nutrition and sepsis. Poorer outcomes amongst outborn infants is likely to be multifactorial and may be related to the period of diagnosis

(postnatal), delays in transport and insufficient expertise with gastroschisis care at the outborn facilities.

Further studies, however, are needed to determine if there is any association between existing transfer protocols and adverse outcomes in patients with gastroschisis within our healthcare system. Improved antenatal care outside of the UHWI, may therefore, improve the outcome of patients with gastroschisis in our country. This study has also highlighted the need for proper transfer protocols to be put in place for cases with gastroschisis in order to improve outcomes.

Sepsis, which occurred in 75 per cent of the cohort, was the significant independent predictor of mortality. These findings are similar to other third world countries. Sekabira *et al* in their study conducted in Africa revealed an overall mortality rate of 43 per cent with sepsis being the leading cause (8). In our study all cases of sepsis were outborn infants, suggesting that lack of proper transfer protocols and lack of immediate paediatric surgical intervention maybe contributing factors for our high mortality.

Limitations of this study are mainly secondary to the retrospective design and the small cohort. Also, our outborn population consists of patients from varied facilities, thus, making this population difficult to assess. There were challenges with the availability of data, mainly missing antenatal records of the mothers for the outborn infants. This posed challenges in evaluating possible antenatal predictors for gastroschisis in this study. The small number of patients made it difficult in determining statistical significance with bivariate analysis. The small cohort also was insufficient for the use of multivariate logistic regression model.

Even with the high probability of survival, infants with gastroschisis still face potential significant long-term morbidity. Our data suggested that adequate antenatal care; antenatal diagnosis and delivery at a tertiary care institution may result in better neonatal outcomes.

CONCLUSION

This study has identified perinatal predictors for gastroschisis outcome with the timing of diagnosis and birthplace proving to be the most significant. The other important factors were birthweight and antenatal ultrasound surveillance. Further detailed studies are needed to investigate antenatal predictors amongst our obstetric population which may provide the potential benefit of being able to risk stratify infants with gastroschisis to allow better prenatal and post-delivery management strategies, overall providing better neonatal outcomes.

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