Original Article

Prevalence of Gastroschisis and its Neonatal Mortality in the Eastern Cape Province Tertiary Institutions

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Abstract

Context: Gastroschisis is a common abdominal wall defect faced by paediatric surgeons worldwide. Early gastroschisis detection, access to improved neonatal intensive care, parenteral nutrition and surgical techniques have led to a reported improvement in mortality of between 4% and 8% in high-income countries. In low to middle income countries, such as in Southern Africa, however, there is as much as 84% mortality among patients with gastroschisis. This is thought to be due to factors such as lack of antenatal screening, access to neonatal intensive care services and parenteral nutrition. **Aims:** The purpose of this study was to calculate the prevalence of gastroschisis and report on its neonatal mortality in the Eastern Cape Province of South Africa. **Settings and Design:** A retrospective observational study on all neonates with gastroschisis, presenting to a tertiary facility offering paediatric surgical services within the Eastern Cape Province from 1 January 2016 to 31 December 2018. **Subjects and Methods:** A convenience sampling method was used in retrieving patient files for the study period. Statistical analysis used: Stata version 13. **Results:** Thirty-seven neonates were included in the study. The prevalence of gastroschisis ranged from 0.07% to 0.18% throughout the 3-year study. The majority (81%) of the neonates were outborn and delivered by mode of caesarean section. Nearly 60% ($n = 22$) were female. 54% ($n = 20$) of neonates died within the neonatal period. **Conclusions:** The majority of the neonates in this study were outborn and female. Although their mortality rate was higher than reported in high-income countries, it was much improved from what is reported in the low to middle income countries.

Keywords: Abdominal wall defect, gastroschisis, neonatal mortality

Introduction

Gastroschisis is one in a spectrum of congenital abdominal wall defects. Typically, the abdominal wall defect lies to the right of the umbilical cord insertion with varying amount of uncovered intestines protruding through the defect. Gastroschisis has fewer associated anomalies, the main anomaly being intestinal atresia, which affects 28% of the neonates.[1]

Gastroschisis can be classified as simple or complex. Simple gastroschisis encompasses abnormally protruding uncovered bowel with no other associated gastrointestinal tract pathology. Complex gastroschisis entails added intestinal pathology such as necrosis, perforation, atresia or volvulus.[2,3] Although gastroschisis has an estimated mortality rate of 4-8% in high-income countries, higher mortality rates of 65% and 84% have been reported in South Africa and Zimbabwe, respectively.[1,4,5]

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The aim of this study was to analyse data on all neonates in the region with gastroschisis (simple/complex) with the objectives of calculating the prevalence of gastroschisis and reporting on the neonatal mortality rate of gastroschisis.

Subjects and Methods

Study design: A retrospective observational study was conducted. A convenience sampling method, where data could be easily collected from nearby treating facilities, was used in retrieving patient files for the study period.

Study population

The study involved all neonates with gastroschisis presenting to the three tertiary facilities offering paediatric surgical

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services within the Eastern Cape Province of South Africa, from 1 January 2016 to 31 December 2018. These facilities all have access to an Intensive Care Unit, Paediatric Surgeon, Anaesthetist, Neonatologist and Parenteral Nutrition. These hospitals together drain a population of 6,522,700 people, with a third being paediatrics.

Ethics and recruitment

Permission was sought from Walter Sisulu University, Eastern Cape Research Committee, Hospital management, and the heads of departments in the respective institutions. Patient information was stored in a password-protected programme on a personal laptop with random numeric assigned for their identities.

Data analysis

Descriptive statistics used include frequency tabulations and summary statistics. The prevalence of gastroschisis was estimated by dividing the number of cases with gastroschisis, by the total number of deliveries recorded in the study sites during the study using the following formula: $\text{Prevalence} = \left(\frac{\text{Gastroshisis}}{\text{All live births}} \times 100,000 \right)$ All live births value was obtained from the South African National yearly statistics registry for the Eastern Cape Province. Case fatality rate was calculated by dividing the number of deaths from gastroschisis over the selected period of time by the number of infants positively diagnosed with the disease $\left(\frac{\text{Demised gastroschisis}}{\text{All gastroschisis}} \times 100 \right)$. Only neonates with gastroschisis who presented to the treating facility in the public sector where factored into these calculations and those who demised at base hospitals prior to transfer or those managed in the private sector being excluded. Fisher's exact test was used to measure the associations between mortality and place of delivery, mode of delivery, foetal birth weight and maternal age on a 2 × 2 table. Univariate and multivariate logistic regression was then formulated to clarify and examine the different levels of influence on mortality of each explanatory variable. The Kaplan–Meier curve was used to estimate the overall survival distribution. Furthermore, adjusted prediction and marginal effect were used to evaluate the probability of mortality based on the patient's characteristics. A $P < 0.05$ was considered as a statistically significant result.

Results

Total number of gastroschisis cases in the Eastern Cape Province from January 2016 to 31 December 2018 was 43. Six cases were excluded as the 28-day mortality could not be assessed due to missing data [Figure 1]. The remaining 37 cases formed the final cohort. Reported prevalence rate was found to be 0.07%–0.18% over the 3-year period (2016-2018).

The majority (81%) of the neonates were outborn and required plastic wrap to cover the eviscerated bowel as an initial management strategy. Although 62% of these infants were born by caesarean section, these were for obstetric reasons and not for gastroschisis itself, in both the inborn and outborn. The mean weight (standard deviation) was 2112 g (475 g). Nearly 60% of the cases were female [Table 1].

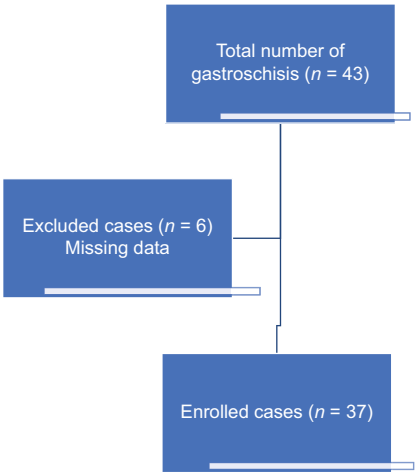


Figure 1: Patient participation

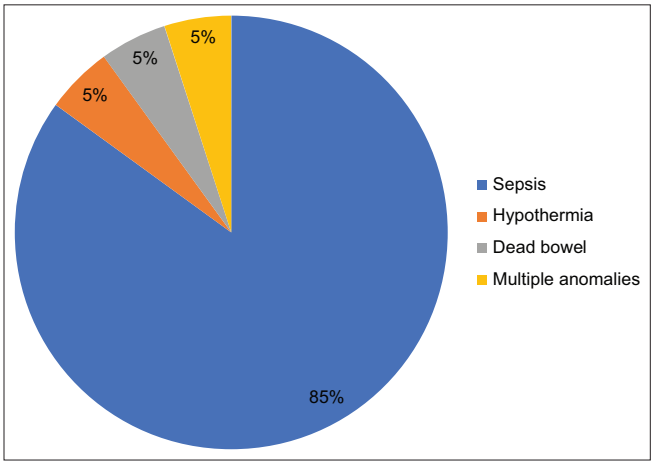


Figure 2: Cause of death

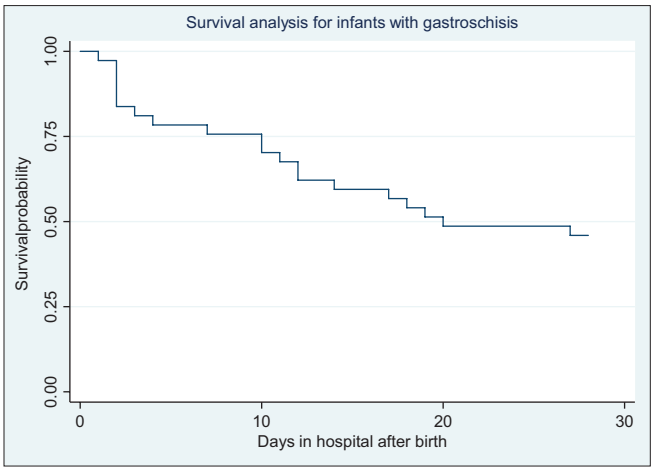


Figure 3: Survival analysis

Table 1: Sociodemographic characteristic of the participant (*n*=37)

Variable	Frequency/percent, *n* (%)
Gender	
Male	15 (41)
Female	22 (59)
Delivery	
Outborn	30 (81)
Inborn	7 (19)
Weight at birth (g)	
1500-2500	28 (76)
2500-3500	9 (24)
Mean (SD) (g)	2112 (475)
Mode of delivery	
NVD	8 (38)
C/S	13 (62)
Maternal age (years)	
15-19	7 (33)
19-29	14 (67)
Mean (SD)	20 (3)
Primary intervention	
Plastic wrap	30 (81)
Silo staged repair	7 (19)
Time until final closure (hours from birth)	
0-24	17 (68)
>24	8 (32)
Duration of hospital stay (days)	
0-28	28 (76)
>28	9 (24)
Mortality	
Died	20 (54)
Alive	17 (46)

NVD: Normal vaginal delivery, C/S: Caesarean section, SD: Standard deviation

Primary fascial closure within 24 h post-delivery occurred in 68% of the neonates. Twelve of the neonates demised before fascial closure was undertaken. As many as 54% ($n = 20$) of the neonates died; the majority the deaths occurred within 20 days post-delivery. Although universal precaution for sepsis with minimal handling of bowel and early initiation of broad-spectrum antibiotics were initiated in all centres, sepsis was still the leading cause of death. As many as 5% of the neonates who demised were hypothermic on admission as shown in Figure 2.

Within this cohort, certain factors were found to adversely contribute to mortality such as being inborn (odds ratio [OR] 1.74), born by caesarean section (OR 1.6) and high maternal age (OR 2.4) while other factors such as high birth weight (OR 0.5) and staged repair of defect (OR 0.4) were seen to translate into a more favourable outcome.

Similarly increased probability of mortality due to being inborn, caesarean section delivery and increased maternal age were noted [Table 2].

Although the Kaplan–Meier curve [Figure 3] depicts survival analysis showing 46% survival at day 28 of life, it can be seen

Table 2: Probability of mortality depending of variables

Variable	Probability	95% CI
Gender		
Male	0.47	0.21-0.71
Female	0.45	0.24-0.66
Delivery		
Outborn	0.43	0.25-0.61
Inborn	0.57	0.20-0.93
Weight at birth (kg)		
≤2.5	0.50	0.30-0.68
>2.5	0.33	0.02-0.64
Mode of delivery		
NVD	0.50	0.15-0.84
C/S	0.61	0.35-0.88
Maternal age (years)		
<19	0.42	0.06-0.79
19-29	0.64	0.39-0.89
Primary intervention		
Plastic wrap	0.50	0.32-0.67
Silo staged repair	0.29	0.04-0.62
Type of closure (h)		
≤24	0.59	0.35-0.82
>24	0.50	0.15-0.84

NVD: Normal vaginal delivery, C/S: Caesarean section, CI: Confidence interval

that the majority of neonates demised within the first 3 weeks of life, with a total of 54% ($n = 20$) mortality within the first 28 days.

Discussion

This study assessed the prevalence of gastroschisis in the Eastern Cape province of South Africa over a 3-year period. A recent South African study reported an apparent increase in the prevalence of 60 cases over a 32-month period compared to a previous report of 48 cases over a 20-year period (1981–2001) by the same group.[4,6] We found the prevalence to be 0.07-0.18% over this period, which is different from the increasing reported rates locally[4] and internationally[7] (from 2.32/10,000 to 4.42/10,000 live births). With the rising prevalence throughout the world,[4,7,8] an increased burden on the health sector, morbidity and mortality, particularly in low to middle income countries, will follow.

There were more females in our cohort, which was similar to other South African studies[4,9] while other studies reported a higher male predominance[3] which demonstrates the heterogeneity of gastroschisis distribution amongst the sexes.

The mortality rate in this study was 54%, which is much lower than reported in low to middle income countries, in spite of the majority of babies being outborn. This was a reassuring finding, especially considering the rural context of the Eastern Cape Province, with travel to some treating facilities being as far as 400 km (±5 h travels time). Unexpectedly, being inborn was found to be a significant risk factor for mortality in our

study. These findings are similar to a study by Sekabira[7] in which 7 of the 13 neonates who had an antenatal diagnosis of gastroschisis and were inborn demised. Our findings are contrary to other studies[10,11] which showed that neonates born in facilities which offered paediatric surgical care (i.e., inborn) and had uncomplicated gastroschisis had the best chance of survival. Due to the majority (81% [n = 30]) of neonates being outborn in this study, a much higher mortality rate than depicted here would be expected in most low to middle income countries.

Within this cohort, the mean maternal age was 20 years, and neonates born to mothers >19 years of age were found to have a higher probability of poor outcome as compared those <19 years of age. This contrasts to what was shown by Rasmussen[12] and Jelena[13] in that young maternal age (<19 years) was deemed to be an increased risk factor to developing gastroschisis and having a poor outcome.

The availability of resources and expertise play an important role in the outcome of gastroschisis and being in a resource-limited region; we were not surprised to see that none of the 37 neonates born within the province during the study were antenatally diagnosed with gastroschisis. This resulted in 81% (n = 30) of the neonates being outborn, with no counselling offered to the mothers, no optimisation of maternal factors which can contribute to early delivery and foetal compromise and delayed referral to the treating institutions.

Most of the deliveries were by caesarean section (62%) due to maternal or foetal indications. In the inborn neonate group, 57% (n = 4) were born by caesarean section and 43% (n = 3) were delivered vaginally. Controversies regarding the best mode of delivery for neonates with gastroschisis are many, with some authors recommending planned caesarean section as it was found to be associated with higher rates of primary closure, shorter duration on mechanical ventilation, earlier establishment of full enteral feeds and shorter hospital stay even in preterm infants,[14] but other studies showed no benefit or even poorer outcome in performing caesarean section for preterm foetus compared to term deliveries.[15] The current literature suggests that routine caesarean section be avoided without an obstetric indication. The probability of mortality due to the selected mode of delivery was 60% higher in caesarean section as compared to those born by vaginal delivery (OR 1.6) in this study. This further reiterates the importance of prenatal diagnosis to assist in planning the mode of delivery and expediting delivery within the treating institution.

Basic management of neonates with gastroschisis entail wrapping the herniated viscera, either with a plastic/cling wrap or a homemade silo from a saline intravenous bag in institutions where preformed silo bags are not freely available. If the neonate is haemodynamically stable, not in respiratory distress and presents early to the facility, the reduction of herniated viscera and primary fascial closure would be attempted. A staged reduction would be done in those who are deemed not suitable candidates for closure with disproportion between the herniated bowel and abdominal cavity. Early primary fascial closure is the treatment of choice for gastroschisis and has been shown to decrease hospital stay total parenteral nutrition dependence and ultimately improve survival.[16] Lin[17] demonstrated that the primary fascial closure was feasible and had added benefits of limiting complications. In this study, the time to intestinal reduction and fascial closure ranged from 6 h up to 13 days. This is attributed to a few factors such as the majority of neonates being outborn and having delays in transportation to the treating facility, paucity of ICU beds to accommodate the neonates and the poor general condition of the neonates on arrival to the facility requiring aggressive resuscitation, which further delayed any early operative intervention.

Plastic bag/cling wrap application was the preferred initial management intervention in this cohort as most centres did not have preformed silos; which was then followed with a surgical intervention. Primary fascial closure was performed within 24 h in 68% of the cases and a staged reduction of bowel with delayed closure in the remaining 32%. As shown by Weinsheimer[16] and Pratheeppanyapat,[18] one tries to achieve fascial closure as soon as possible to establishing intestinal function, limit sepsis and subsequent diminished time to discharge. Primary fascial closure was the preferred choice of management in the 2011 European Paediatric Surgeons' Association Congress survey as it was found to have a good outcome. Even though the majority of neonates in this cohort had primary fascial closure within 24 h, it was found that those who underwent a staged reduction with delayed fascial closure had favourable outcome with mortality odds ratio of 0.7.

Neonates who were deemed not fit for surgical intervention or had a complex gastroschisis (with atretic or necrotic bowel), 32% (n = 12), only had plastic/cling wrap applied with no definitive closure and subsequently demised.

Conclusions

The prevalence of gastroschisis in the Eastern Cape Province was lower than reported locally and internationally. Although eight in ten neonates were outborn, nearly half of them survived. The leading cause of death was sepsis.

Limitations

The small size posed a challenge in adding adequate power to the study. Incomplete and missing medical data from the patient's records hampered data analysis. The exclusion of neonates with gastroschisis managed in the private sector further decreased the sample size and study power. The use of case fatality rate instead of neonatal mortality rate was deemed more feasible in this study.

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Conflicts of interest

There are no conflicts of interest.

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