

Original Research Article

AN INVESTIGATION INTO THE VARIABLES THAT IMPROVE OUTCOMES IN NEONATES WITH GASTROSCHISIS IN A TERTIARY CARE FACILITY

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ABSTRACT

Background: Gastroschisis is a congenital anterior abdominal wall defect in which the abdominal contents protrude out of the abdomen. The condition requires urgent surgical intervention. The study aimed to evaluate the factors influencing outcomes in gastroschisis.

Materials and Methods: A retrospective study was conducted in which data from neonates with gastroschisis managed at our tertiary care centre between September 2019 and June 2022 were analysed. Data on multiple obstetric, perinatal, preoperative, and surgical factors were collected, and their effects on overall survival were analysed.

Results: There were 26 male and 12 female patients, out of which 47.3% were preterm neonates and 78.9% had birth weight less than 2.5 kg. Antenatal scans were performed in 26% of cases and were normal. Out of 30 hospital-delivered children, only one was an inborn child, and the others were referred. Seven neonates were home-born. All referred neonates were brought in poor general condition. 36.8% patients had near normal bowel with 71.4% survival, and the rest were matted, pregangrenous or gangrenous, with survival of only one patient. 73.7% of patients underwent abdominal wall closure, while 26.3% underwent staged reduction with silo bag application. The overall survival was 29%, but none of the patients with a silo bag survived. The mean time of death was 35.7 hours, and the mean time to discharge was 14.4 days.

Conclusion: Overall survival rate in our study was 29%. The presence of edematous bowel with matting and patients requiring silo bag were significantly associated with mortality.

Keywords: Gastroschisis, Neonate, low-middle-income countries, Survival.

INTRODUCTION

Gastroschisis (GS) is a defect of the anterior abdominal wall, usually on the right side, with fetal viscera (intestines, stomach) into the amniotic space and requires immediate treatment after birth. [1,2] The incidence of GS is rising and currently is 1 in 4,000 live births. [3,4,5] Although there is a strong association with young maternal age, the aetiology of GS is unknown in most cases. [6] In developed countries, it is invariably diagnosed antenatally, and the survival rate is over 90%. [7] However, in low and middle-income countries (LMIC), survival rates vary from 0 to 45%. [8,9,10] These defects are usually diagnosed

postnatally at most of the primary healthcare centres and then referred to tertiary institutes. Delays in appropriate neonatal surgical care and inadequate pre-hospital management result in many neonates presenting with compromised fluid and thermodynamics, coagulopathy, sepsis, and poor bowel conditions like contamination or damage to the vascular pedicle, resulting in intestinal ischemia and necrosis.^[9-11]

Several other factors, like antenatal diagnosis, mode of delivery, place of delivery, gestational age, type and timing of abdominal wall closure, necrotising enterocolitis (NEC), and associated malformations like intestinal atresia, influence the outcome of GS_{.[12,13]}

The most common surgical treatment is primary closure under general anaesthesia or serial reductions using a preformed silo bag over several days, followed by abdominal wall closure. [14,15]

The poor survival rates make it difficult to study prognostic predictors of gastroschisis. We have seen improved outcomes and survival rates in recent years among patients with Gastroschisis at our centre. Therefore, the present study was done to identify the factors that favour survival and improve outcomes.

MATERIALS AND METHODS

Study setting, Design and duration: A retrospective study was conducted at our tertiary care centre between September 2019 and June 2022, and data from neonates with gastroschisis were analysed. Data were collected from patients' databases and medical records available at our centre. Data on antenatal and postnatal history, referral, condition at admission, surgical intervention, and post-surgical management were retrieved, and their effects on survival were analysed.

Preoperative management: On admission to the NICU, vitals were recorded, and intravenous fluids and prophylactic broad-spectrum intravenous antibiotics were administered. Care was taken to maintain a warm environment, keeping the baby dry and preventing heat loss. The patients were kept nil per oral with nasogastric decompression and per urethral catheterisation. No forced attempts were made to reduce the bowel bedside. The herniated bowel and contents were enclosed with a warm saline-soaked sterile gauze piece. The baby was shifted to the operating room as early as possible.

Principles of surgical management: After general anesthesia, a thorough evaluation of the bowel was done. Eviscerated contents were washed, decompressed, and reduced whenever possible through the primary defect. Abdominal wall closure, with or without fascial closure, was performed using interrupted sutures.

In cases where the bowel was thickened and edematous, and bowel reduction could not be done in a tension-free manner, silo bag application was done. A sterile urobag was cut to the appropriate width and sutured to create a silo. Bowel loops were placed inside it, and the edges of the silo were sutured to the margins of the abdominal wall defect with continuous locking silk sutures for a watertight closure. The distal end of the silo was tied with a bandage piece and hung from an overhead supporting beam. Serial

reduction was done daily, and a delayed abdominal wall closure was attempted in 7–14 days.

Postoperative management: Aggressive postoperative monitoring and fluid resuscitation were done. The majority of patients received higher antibiotics. A few of the patients with primary abdominal closure required ventilatory care, and all the patients of silo bag application were electively kept on ventilator support and sedation. Bowel decompression was done by nasogastric aspiration, and daily per rectal suppository till the abdomen was soft. The bladder was also kept decompressed with a catheter. Either partial or total parental nutrition (TPN) was initiated. Enteral feeds were started once the bowel function started and nasogastric aspirates decreased. Expressed breastmilk feeding via an infant feeding tube was encouraged.

Outcome Variables: The endpoint of the study was death or hospital discharge. Outcome was evaluated in terms of survival following management of gastroschisis and percentage of survivors of gastroschisis in terms of gestational age, birth weight, mode of delivery, centre of delivery, time of surgery, surgical technique of closure, associated anomalies, need for postoperative ventilation, neonatal intensive care unit (NICU) requirement, and time to full feeds. Data Analysis: All medical records were carefully recorded in Microsoft Excel spreadsheets, and data analysis was performed using Microsoft Excel and Jamovi.

RESULTS

A total of 38 neonates with a male-to-female ratio of 1.9:1 were admitted to our department. There were 18 (47.36%) pre-term and 20 (53.64%) term neonates. 8 (21.05%) were very low birth weight (VLBW), 22 (57.8%) were low birth weight (LBW), and 8 (21.05%) were more than 2.5 kg weight. While the majority of newborns 29 (42.6%) were referred, only one newborn was delivered at our centre, and 7 (18.4%) were delivered at home. It was noted that 10 (26.4%) of the newborns were admitted within 6 hours of birth, and 18 (47.7%) within 24 hours. However, 9 (23.3%) neonates presented to the hospital beyond 24 hours after birth. The antenatal scan was normal in 10 (26%) patients, and the rest did not undergo any scans. The mode of delivery was vaginal in 30 (79.6%) patients, and 7 (18.4%) were delivered via caesarean section. There was one patient for whom these details were unavailable, as the baby was found abandoned in the bushes at a distant village and then brought to our centre for treatment.

Table 1: Obstetric and perinatal factors a	ffecting the outcome o	of GS		
	Survival (n=11) n (%)	Expired (n=27) n (%)	Total (n=38) n (%)	p- value
Age of gestation				0.11
Preterm	3(16.66%)	15(83.34%)	18 (47.36%)	
Full term	8 (40%)	12 (60%)	20 (53.64%)	

Gender				0.27
• Male	6 (23%)	20 (77%)	26 (68.4%)	
 Female 	5 (41.6%)	7 (58.3%)	12 (31.6%)	
Birth weight				0.25
• <1.5 kg	1 (12.5%)	7 (87.5%)	8 (21.1%)	
• 1.5-2.5 kg	6 (27.2%)	16 (72.7%)	22 (57.8%)	
• >2.5 kg	4 (50%)	4 (50%)	8 (21.1%)	
Place of delivery				0.06
 Institutional 	10 (33.3%)	20 (66.6%)	30 (78.9%)	
 Inborn 	1 (100%)	0 (0%)	1	
 Outborn 	9 (31.03%)	20 (69.1%)	29	
 Home 	0 (0)	7 (100%)	7 (18.4%)	
 Unknown 	1 (1)	0 (0)	1 (2.6%)	
Mode of delivery				0.17
 Normal vaginal delivery 	7 (23.3%)	23 (77.7%)	30 (79.6%)	
Cesarean section	3 (43.3%)	4 (57.7%)	7 (18.4%)	
 Unknown 	1 (100%)	0 (0)	1 (2.6%)	
Age at admission to our hospital				0.08
• <6 hours	4 (40%)	6 (60%)	10 (26.4%)	
• 6-24 hours	6 (33.3%)	12 (66.6%)	18 (47.7%)	
• >24 hours	0 (0)	9 (100%)	9 (23.3%)	
 unknown 	1 (100%)	0 (0)	1 (2.6%)	
ANC USG				0.73
 Done and Normal 	2 (20.0%)	8 (80.0%)	10 (26.0%)	
 Done and diagnosed 	0	0	0	
Not done	8 (33.3%)	16 (66.6%)	24 (63.0%)	
 Unknown 	1 (25.0%)	3 (75.0%)	4 (10.0%)	

All the referred patients were brought in poor general condition and without proper protection of the eviscerated bowel. At admission, 14 (36.8%) patients had near normal-looking bowel, 7 (18.4%) had edematous, matted bowel with adhesions, 16 (42.0%)

had pregangrenous changes, and 1 (2.6%) had gangrene. A few patients had other associated anomalies, like cardiac anomaly in 2, intestinal atresia in 2, malrotation of the gut in 5, and anorectal malformation in 1 patient.

Table 2: Preoperative and surgical factors affecting the outcome of GS

Preoperative and surgical factors	Survival	Expired (n=27)	Total (n=38) n (%)	P value	
	(n=11)				
	n (%)	n (%)			
Condition of bowel at admission					
Normal bowel/edematous non matted	10 (71.4%)	4 (29.6%)	14 (36.8%)		
Edematous matted bowel with		, , ,	, ,		
adhesions	1 (6.0%)	15 (94.0%)	16 (42.0%)		
 adhesions + pregangrenous 	0	7 (100%)	7 (18.4%)		
 adhesions + gangrenous 	0	1 (100%)	1 (2.6%)		
Associated anomalies adding comorbidity					
Cardiac	0	2 (100%)	2 (5.3%)		
Malrotation of the gut	1 (20%)	4 (80%)	5 (13.2%)		
Anorectal malformation	0	1 (100%)	1 (2.6%)		
Intestinal atresia	0	2 (100%)	2 (5.3%)		
Surgical intervention					
Abdominal wall closure	11 (39.9%)	17 (60.7%)	28 (73.7%)		
Silo bag application	0	10 (100%)	10 (26.3%)		
Additional procedure:					
• Stomy	0	2	2		
 Resection anastomosis 	0	1	1		

Abdominal wall closure was done in 28 (73.7%) patients, while silo bag application was done in 10 (26.3%) patients. Additionally, one patient with bowel atresia and 1 with gangrenous bowel required stomy formation, and one patient with atresia required bowel anastomosis. Only one patient in the silo bag survived to undergo abdominal wall closure later. Postoperatively, all neonates with a silo bag and 18 with abdominal wall closure required ventilatory care. Partial TPN with essential amino acids at 1-2 g/kg was initiated in all patients once they were stable. The mean time to start enteral feeds was 6.6 days, and the mean time for full feeds was 10 days.

The mean time to discharge was 14.4 days (10-26 days).





Figure 1: Post-surgical case of GS (a) stitch line after abdominal wall closure, (b) silo bag application

The overall survival rate was 29% with no survival in the silo bag procedure group. Overall, the mean time of death was 35.7 hours, while the mean time of death in the anatomical closure group was 41 hours, and in the silo bag procedure group was 19 hours.

It is observed that the condition of the bowel at admission and the surgical procedure performed are significantly correlated with survival. However, other factors, such as gestational age, gender, birth weight, place and mode of delivery, antenatal diagnosis, and associated co-morbid congenital conditions, do not show any significant correlation with survival in neonates with gastroschisis.

DISCUSSION

GS is a curable congenital anterior wall anomaly, which, when addressed early after birth, has a good prognosis.^[9,13] In our study, a male predominance was seen, similar to studies from other LMICs and high-income countries (HICs).^[16-20]

The proportion of preterm newborns in our series was slightly lower than that of term babies (47.36% vs 53.64%), in contrast to other studies. [13,17] The deleterious effects of prematurity, like lung immaturity and the need for mechanical ventilation, an increased risk of sepsis, delayed onset of bowel movements and feeding, negatively affect the prognosis of gastroschisis. Hence, the survival of preterm neonates was less (16.6%) and is described by many investigators, even in developed countries. [13,16,21]

The proportion of VLBW and LBW neonates in our study was 77.9%. Survival was better (10 cases) when birth weight was greater than 1.5 kg. Similar findings were seen in other studies as well.^[16] GS patients tend to be small for gestational age and hence are prone to sepsis and hypothermia, need a longer time to start oral intake, have a higher risk of NEC, and longer duration of hospitalization.^[22,23]

Patients who were delivered in the hospital and those who were hemodynamically stable on admission had better survival. The one inborn patient who survived was a preterm, had received immediate resuscitation and stabilisation, and was operated on within a few hours of delivery. Outborn patients (69.1%) who are referred to our centre had not received adequate stabilisation and bowel care, even though health workers often accompany them. Similarly, homedelivered newborns (10.3%) were initially taken to the local hospital and then referred. In such circumstances, the newborns are more susceptible to hypothermia, hypovolemia, sepsis, bowel oedema, and delayed surgery, consequently worsening the prognosis.[16,24,25] This was observed in various studies from India and other LMICs. Studies from the HICs show survival rates of over 90% among inborn patients, whereas in LMICs, most patients are referred after delivery or are home-born, further highlighting the importance of delivery at tertiary centres.[26-31]

Although a few antenatal scans were performed, they were essentially normal. The majority of patients do not seek timely antenatal visits and scans, and are diagnosed soon after birth. This is consistent with findings from other parts of India and LMICs. Antenatal diagnosis of GS, as in the HICs, would give an insight to parents and health workers to seek necessary surgical care at birth and hence, would decrease morbidity and mortality. [31,32,33] In addition, preventive measures for preterm labour, including tocolytics and corticoid therapy, are not possible until gastroschisis is recognised in the antenatal period.

Three-fourths of our patients were delivered vaginally. However, the mode of delivery was not found to be associated with survival in our study or in other studies, and further studies are required to establish any guidelines.^[16,34,35]

Several congenital anomalies may be associated with GS in up to 30% cases.^[7,36] Conditions like congenital cardiac anomalies, trisomy,^[18] bowel atresia, malrotation, and anorectal malformation increase the morbidity and mortality. Complex GS is a term used when additional bowel damage is present. All patients with comorbid congenital anomalies and complex GS, which were managed at the time of surgery, did not survive.

GS usually presents with small, underdeveloped abdominal cavities, and the eviscerated, uncovered bowel is in continuous contact with the external environment, which makes it inflamed, edematous, thickened, and often matted, with overlying peel. The extent of bowel oedema and matting is directly proportional to the time interval between birth and surgical intervention. Such a situation can make it difficult to reduce contents into the abdominal cavity or to reduce them at all.[37,38,39] In our series, 36.8% of patients had near-normal bowel with 71.4% survival, and the rest were matted, pregangrenous, or gangrenous, with survival of only one patient with matted bowel. This finding was statistically significant (p-value 0.0002), and similar findings were observed in other studies.^[17]

Surgical management of GS depends on the general condition of the neonate, the eviscerated contents, and the anticipated level of abdominal pressure after closure.[37] The closure is either primary or a staged closure. Primary reduction with sutured fascial defect closure is the preferred standard operative procedure.[32,33] When the fascial defect cannot be closed, only skin closure is done. Operative staged reduction using a silo bag to the enlarged defect and delayed defect closure is considered when bowel cannot be reduced completely and when there is a risk of abdominal compartment syndrome. [32] In this study, abdominal wall closure was done in threequarters of patients, while silo bag application was done in fewer patients. The survival in the first group is statistically significant compared to the second group, and similar findings were seen in other studies from India and other nations. [17,21,40]

The overall survival of gastroschisis in our study was 29%, which is comparable with survival reported

from many LMICs, which range from 0% to 45%.[8,9,10] Certain Indian studies report even higher survival rates of 44.83% and 72.41%.[16,17] The difference in our survival rate can be attributed to the fact that, despite having a tertiary-level neonatal care unit and all the necessary infrastructure, the patients of GS brought to us are mostly from the lower socioeconomic class and arrive in poor general condition, with grossly contaminated bowel and sepsis, and inadequate neonatal resuscitation at peripheral centres. Many times, they come from long distances (Rajasthan, Madhya Pradesh). Also, at the hospital, surgery is sometimes delayed, and one-toone nursing care is not possible due to the busy and over-occupied NICU. Factors such as gestational age, gender, birth weight, place and mode of delivery, antenatal diagnosis, and associated comorbid congenital conditions do not show a statistically significant correlation with survival in our study, in contrast to many other studies.^[16,17,21] Limitations of the study included the inability to evaluate long-term outcomes, as most patients did not attend proper follow-up.

CONCLUSION

Our study identifies factors that affect the outcomes of babies with GS, but survival remains dismal. GS is often diagnosed antenatally, and since the uterus remains the ideal and most economical transportation unit, the neonates should be delivered in a centre where surgery can be performed immediately after the birth. Hence, antenatal check-up and measures to delay preterm delivery should be encouraged. When delivered at a peripheral hospital, the bowel can be kept in a sterile urobag, and then the child transferred as early as possible. Abdominal wall closure of GS within a few hours of birth will reduce the burden of the 2-stage procedure and sepsis and hence increase survival. Aggressive management of the child after admission, good nursing care, use of higher-dose antibiotics, enteral and parenteral nutrition, and sepsis control will further improve survival. Knowledge and education about conditions like gastroschisis, their management, and survival for the public are also necessary.

Abbreviations

Gastroschisis (GS)

Low and middle-income countries (LMIC)

Necrotising enterocolitis (NEC)

Neonatal intensive care unit (NICU)

Total parental nutrition (TPN)

Very low birth weight (VLBW)

Low birth weight (LBW)

High-income countries (HICs)

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