Anterior abdominal wall defects: Demographic and clinical profile and outcome at a tertiary care hospital

Parveen Kumar, Vivek Manchanda,* Mamta Sengar
Department of Pediatric Surgery, Chacha Nehru Bal Chikitsalaya, New Delhi, India

Correspondence*: Dr. Vivek Manchanda, Associate Professor, Department of Surgery Chacha Nehru Bal Chikitsalaya, New Delhi, India. E-mail: vivek_7477@hotmail.com

KEYWORDS
Primary closure, Length of stay, Mortality, Neonate

ABSTRACT
Background: The anterior abdominal defects, especially gastroschisis and omphalocele have high mortality rates in developing countries. Time to intervene has been hypothesized to be associated with morbidity and mortality. The aim was to determine factors affecting mortality in neonates with anterior abdominal wall defects.

Methods: This retrospective descriptive study was done at a tertiary pediatric care center. The medical records of patients with a diagnosis of anterior abdominal defects (omphalocele/gastroschisis/umbilical cord hernia) admitted at our center from Jan 2015 to Dec 2019 were retrieved. The demographic and clinical data were studied including age, sex, religion, gestational age, associated anomalies, electrolytes at admission, septic profile, operative details, length of hospital stay, and mortality. The statistical data was fed on a Microsoft Excel worksheet and analysis was done.

Results: Thirty-nine (39) neonates were included in the study. M:F ratio was 2:1, with 61.5% belonging to the Hindu religion. The majority had term gestation (87.2%). Birth weight ranged from 1.5 to 4 kg (mean 2.47 ±0.5 kg). Eighteen (18) neonates had gastroschisis, 15 omphalocele major and 6 omphalocele minor, with a median age of presentation at 1 day of life. Time to intervention ranged from 0 to 5 days (interquartile range 1-1.25 days) after admission. Primary closure could be achieved in the majority (66.7%), while ventral hernia was created in 17.9% and the silo was needed in the rest. The mean postoperative length of stay was 9.31 days (±9.85 days) with a survival rate of 38.5%. The mortality rate in gastroschisis and omphaloceles were 61.1% and 38.1% respectively. The significant factors for survival were birth weight, and primary abdominal wall closure.

Conclusions: The present study brings out a different clinical profile of anterior abdominal wall defect patients. We recommend early surgery soon after stabilization and primary abdominal wall repair whenever abdominal pressures permit.

INTRODUCTION
The gastroschisis and omphalocele account for the most common anterior abdominal wall defects, with an incidence of 1:12000 and 1:4000 live births, respectively.[1,2] In gastroschisis, usually small intestine, occasionally stomach or colon lies outside body cavity without any protective sac or membranes. While in omphalocele, a thin membrane surrounds the protruding organs that may include the liver, small intestine, spleen, stomach, or gonads.[3] Gastroschisis is rarely associated with non-intestinal anomalies, but intestinal atresias are found in up to 28% of cases. On the contrary, non-intestinal anomalies, including cardiac defects and pulmonary hypoplasia, are reported in up to 24% to 50% of omphalocele cases.[3-7] The overall survival in gastroschisis is 92–96% in the developed world and prognosis is determined by the degree of injury to the bowel, the severity of illness in the first week of life, and the immediate post-operative recovery.[8,9] The mortality in omphalocele is higher (20%) and may be related to other anomalies including intestinal atresia, cardiac defects, or pulmonary hypoplasia.[3,10] These anomalies and their severity affect survival rates for live-born children with omphalocele.[11] We did this retrospective study to estimate the mortality rate in neonates with anterior abdominal wall defects and whether there is any difference in the same as compared to various other western studies. Also, we
aim to study factors affecting mortality in these patients.

METHODS

It was a retrospective study (descriptive) done at the pediatric surgery department of a tertiary care hospital after taking IRB approval. The hospital medical records from Jan 2015 to Dec 2019 were retrieved to find out cases of anterior abdominal wall defects (omphalocele/gastroschisis/umbilical cord hernia). All neonates presenting with anterior abdominal wall defects were included in the study, while those presenting late after escharization were excluded from the study. The demographic and clinical profile of patients were studied including age, sex, religion, gestational age, birth weight, associated anomalies, age at presentation, electrolytes at admission, septic profile, duration between admission and surgery (time to intervene), operative details, length of hospital stay and mortality. The data was fed on a Microsoft Excel worksheet and analysis was performed. Mean, median, and the range were calculated for continuous variables. Student t-Test and chi-square test were used for statistical analysis and a p-value < 0.05 was considered significant.

Management Protocol: Our institute is a referral pediatric center with no facilities for labor. Hence, all patients are outborn. In our institution, all ventral abdominal defects are resuscitated at admission. When stable, patients are taken for primary closure, when feasible, or escharization, when primary closure is not feasible with intact sac. If the sac is ruptured or there is a complicated ventral abdominal wall defect or gastroschisis not amenable to primary closure, operative silo creation is resorted to, and staged closure is done.

RESULTS

A total of 41 medical records of patients operated on for anterior abdominal wall defect were retrieved. Two infants with post escharization of sac were excluded from the study and 39 neonates were included. The demographic and clinical details are mentioned in Table 1. Only 2 neonates had positive blood culture sent at admission and received antibiotics as per sensitivity pattern. Thirty-six out of 39 neonates had signs of dehydration at presentation and 28 had abnormal electrolytes, while all received fluid resuscitation at admission. The survival findings are presented in Table 2. Four patients who went home against medical advice were considered mortality for analysis as the condition at discharge was not compatible with the life.

The mortality rate in gastroschisis and omphalocoeles were 61.1% and 38.1%, respectively, with an overall survival rate of 38.5%. The duration between admission and surgery (time to intervene) was not found to be significantly associated with mortality, though half of the neonates operated the next day after admission survived while 5 (71.5%) among the 7 operated immediately after admission expired. The birth weight of >2.6 kg was associated with significant overall survival (p=0.02). There was no statistical significance in mortality among various types of anterior abdominal wall defects. The primary abdominal wall closure was associated with significant overall survival (p=0.01). Postoperative hospital stay was significantly high in the survival group (p=0.002).

DISCUSSION

The antenatal diagnosis of anterior abdominal wall defects has seen an upsurge. It may be due to better equipment and expertise of radiologists over time. It provides the benefit of parental counseling and helps in planning the delivery of the baby. It is suggested that the delivery be planned at a center with neonatal surgery facility and early repositioning of the bowel before bowel edema or injury sets in. However, the same has not been seen in developing countries. It is even more important in omphalocoele, which is associated with frequent concomitant abnormalities. Goetzinger et al. studied sonographic predictors of postnatal bowel atresia in fetal gastroschisis and found that intra-abdominal bowel dilation >14mm was associated with an increased risk for postnatal bowel atresia in fetal gastroschisis.[12] The survival in omphalocoeles depends on associated anomalies.[11,13] The large defects in omphalocoele are usually managed conservatively, allowing the sac to epithelialize and postponing surgery till later in life.[14] During the study period, we had 2 infants post-escharization or epithelization of the sac and successfully underwent adhesiolysis and abdominal wall closure.

As per our hospital policy, primary abdominal wall closure was attempted in all patients with gastroschisis/omphalocoele. If intra-abdominal pressure was high at attempted primary closure, only skin cover (ventral hernia creation) was attempted. If high abdominal pressure precluded it, silo creation was attempted followed by staged closure.

There was male preponderance in our study, similar to the study by Bucher et al and Erdogan et al.[15,16] Roessingh et al. studied 68 cases of gastroschisis retrospectively from 2000 to 2007 and found that 53% were females.[17] In our study, 16 out of 18 gastroschisis (88.9%) were simple gastroschisis. Two gastroschisis patients (11.1%) had associated anomalies (microcephaly and hydrocephalus) while 8 omphalocoele patients (38%) had associated anomalies (cardiac and hydrocephalus most common). Schmedding et al. reported similar
results with 82% of gastroschisis as simple and mainly associated anomalies in omphalocele were trisomies (18 and 21), cardiac anomalies, and urinary tract anomalies.[18] Our non-GI anomaly rate in gastroschisis was similar to that reported by Payne et al.[15,19]

Table 1: Demographic and clinical profile of patients operated for anterior abdominal wall defects

| Parameter                  | N   | Percentage | p value |
|----------------------------|-----|------------|---------|
| Gender                     |     |            | 0.26    |
| Male                       | 26  | 66.7%      |         |
| Female                     | 13  | 33.3%      |         |
| Religion                   |     |            |         |
| Hindu                      | 24  | 61.5%      |         |
| Muslim                     | 15  | 38.5%      |         |
| Gestational Age            |     |            | 0.13    |
| Term                       | 34  | 87.2%      |         |
| Pre-term                   | 5   | 12.8%      |         |
| Weight                     |     |            | <0.001  |
| Mean                       | 2.47 kg |         |         |
| Standard deviation         | 0.50 kg |         |         |
| Type of Defect             |     |            |         |
| Gastroschisis              | 18  | 46.2%      |         |
| Omphalocele Major          | 15  | 38.5%      |         |
| Omphalocele Minor          | 6   | 15.4%      |         |
| Age at presentation        |     |            |         |
| Min                        | 0 day |         |         |
| Max                        | 13 days |        |         |
| Median                     | 1 day |         |         |
| Interquartile range        | 1-2 days |       |         |
| Procedure Done             |     |            |         |
| Primary Closure            | 26  | 66.7%      |         |
| Silo Formation             | 6   | 15.4%      |         |
| Ventral Hernia             | 7   | 17.9%      |         |
| Days Waiting for Surgery   |     |            | 0.0003  |
| Min                        | 0 day |         |         |
| Max                        | 5 days |        |         |
| Median                     | 1 day |         |         |
| Interquartile range        | 1-1.25 days |       |         |
| Length of Stay after Surgery|    |            |         |
| Min                        | 0 day |         |         |
| Max                        | 43 days |        |         |
| Median                     | 7 days |         |         |
| Interquartile range        | 2 - 10 days |       |         |
| Outcome                    |     |            |         |
| Survived                   | 15  | 38.5%      |         |
| Expired                    | 20  | 51.3%      |         |
| LAMA                       | 4   | 10.3%      |         |
Table 2: Survival Statistics

| Birth Weight | Survived | Expired | Total | p value |
|--------------|----------|---------|-------|---------|
| ≤ 2.6 kg    | 10       | 16      | 26    | 0.02    |
| >2.6 kg     | 10       | 3       | 13    |         |
| Total       | 20       | 19      | 39    |         |

| Type of Defect | Survived | Expired | Total | p value |
|---------------|----------|---------|-------|---------|
| Gastroscisis  | 7        | 11      | 18    | 0.17    |
| Omphalocele   | 13       | 8       | 21    |         |
| Total         | 20       | 19      | 39    |         |

| Surgery Done | Survived | Expired | Total | p value |
|--------------|----------|---------|-------|---------|
| Primary Closure | 17   | 9       | 26    | 0.01    |
| Silo Formation | 0     | 6       | 6     |         |
| Ventral Hernia | 3     | 4       | 7     |         |
| Total         | 20      | 19      | 39    |         |

| Time to Surgery | Survived | Expired | Total | p value |
|----------------|----------|---------|-------|---------|
| 0 days         | 2        | 5       | 7     | 0.36    |
| 1 day          | 11       | 11      | 22    |         |
| 2 days         | 4        | 2       | 6     |         |
| >2 days        | 3        | 1       | 4     |         |
| Total          | 20       | 19      | 39    |         |

| Length of Stay | Survived | Expired | Total | p value |
|----------------|----------|---------|-------|---------|
| <7 days        | 5        | 14      | 19    | 0.002   |
| ≥7 days        | 15       | 5       | 20    |         |
| Total          | 20       | 19      | 39    |         |

We considered LAMA (left against medical advice) patients as potential mortality as it is not possible for a neonate to survive at home without achieving full feeds at the hospital especially in poor socioeconomic strata and limited access to nutrition and medical care at remote places. There was a huge difference in our mortality rates as compared to various other studies.[15-24] These differences might be due to various demographic and environmental factors in middle-income countries like India. Wright et al. emphasized the role of surgical intervention in the survival of gastroscisis and documented a high mortality rate with contrasting statistics. It has 75% to 100% mortality in low middle-income countries while it is <4% in high-income countries.[24] In our study, the majority of mortality in both groups happened within 7 days of operation. Schmedding et al. also reported that half of gastroscisis mortality patients and 44% of omphalocele mortality patients expired in the first 10 days of life.[18]

The significant factors for survival in our patients were birth weight >2.6 kg (p=0.02) and primary abdominal wall closure (p=0.01). It is not in concordance with the developed world where birth weight above 1500 grams is associated with better survival.[18] This might be explained with better availability of intensive care, mechanical ventilation, parental nutrition, and medical resources in developed nations. High mortality rates in our study also explain the lesser median length of hospital stay (7 days) as compared to 33 days, 33 days, and 55 days as studied by Melov et al., Payne et al., and Bucher et al.[15,19,22]

Traditionally, omphalocele had been classified as major and minor variants depending on the size of the defect. The omphalocele minor was labeled with defect size ≤4 cm. Though this distinction is not being followed currently. Kumar et al. studied the impact of omphalocele size on associated anomalies and found that small omphalocele size (≤4 cm) correlates with an increased prevalence of associated gastrointestinal anomalies, a lower prevalence of cardiac anomalies, and a higher predominance of male sex (P = 0.01).[25] In our study, all 6 patients with omphalocele minor were incidentally male, none had cardiac anomalies or gastrointestinal anomalies.

A meta-analysis by Kunz et al. showed that silo closure was associated with better clinical outcomes in the studies with the least selection bias, but when all studies were included, primary closure was associated with improved survival.[26] In our study also, primary closure was associated with improved survival. However, all 6 patients who underwent silo creation expired. We can relate this to sepsis and inadequate support in form of parenteral nutrition in these babies.

The mean birth weight in our study was 2.47 ±0.5 kg, similar to Payne et al., Watanabe et al., and Overcash et al.[19-21] The gestational age as reported by Payne et al. was 36 ±2 weeks, while Erdogan et al. reported 14 preterm gestations out of 29 gastroscisis neonates.[16,19] We, however, have mostly term neonates in our series. We had 5 preterm neonates in our study including 4 with gastroscisis. The low number of preterm neonates in our study may be due to the fact that our institute does not have any inborn neonates. Thirty-six out of 39 neonates had signs of dehydration at presentation and 28 had abnormal electrolytes (hyponatremic dehydration). All neonates received fluid resuscitation at admission. We did not see any significant effect of dyselectrolytemia at the presentation on survival.

In our study, 10 out of 18 gastroscisis (55.5%) and 16 out of 21 (76.2%) omphaloceles could achieve complete primary closure. These closure rates were in accordance with other studies in the literature.[19-23] The overall primary closure rate in our patients (66.7%) was less as compared to the study by Schmedding et al. (73%). Due to the limited availability of postoperative mechanical ventilation at our center, neonates with borderline abdominal pressures had to be managed by a ventral hernia or silo creation rather than complete primary closure.

The mean birth weight in our study was 2.47 ±0.5 kg, similar to Payne et al., Watanabe et al., and Overcash et al.[19-21] The gestational age as reported by Payne et al. was 36 ±2 weeks, while Erdogan et al. reported 14 preterm gestations out of 29 gastroscisis neonates.[16,19] We, however, have mostly term neonates in our series. We had 5 preterm neonates in our study including 4 with gastroscisis. The low number of preterm neonates in our study may be due to the fact that our institute does not have any inborn neonates. Thirty-six out of 39 neonates had signs of dehydration at presentation and 28 had abnormal electrolytes (hyponatremic dehydration). All neonates received fluid resuscitation at admission. We did not see any significant effect of dyselectrolytemia at the presentation on survival.

In our study, 10 out of 18 gastroscisis (55.5%) and 16 out of 21 (76.2%) omphaloceles could achieve complete primary closure. These closure rates were in accordance with other studies in the literature.[19-23] The overall primary closure rate in our patients (66.7%) was less as compared to the study by Schmedding et al. (73%). Due to the limited availability of postoperative mechanical ventilation at our center, neonates with borderline abdominal pressures had to be managed by a ventral hernia or silo creation rather than complete primary closure.
The limitations of the study include the retrospective design. Our center is a purely children’s hospital, thus selection bias in form of including only outborns with no benefit of antenatal diagnosis or early surgery being provided to the babies. Although only 2 children had positive blood cultures at admission, there are likely high infection rates missed by investigations at admission.

CONCLUSION

The present study brings out a different clinical profile of our subset of anterior abdominal wall defect patients in comparison to the Western world. We found that the birth weight, and primary abdominal wall repair were the factors affecting survival in neonates with ventral abdominal wall defects. We recommend early surgery soon after stabilization and primary abdominal wall repair whenever abdominal pressures permit.

Acknowledgements: Nil

Conflict of Interest: Author (PK) is part of the editorial team; however the manuscript was independently handled by another editor and PK was not involved in the decision making of the manuscript.

Source of Support: Nil

Consent to Publication: No clinical figure is used in the manuscript.

Author Contributions: Author(s) declared to fulfil authorship criteria as devised by ICMJE and approved the final version.

REFERENCES

1. Laughon M, Meyer R, Bose C, Wall A, Otero E, Heereen A, et al. Rising birth prevalence of gastrochisis. J Perinatol. 2003; 23:291-3.
2. Kilby MD. The incidence of gastrochisis. BMJ. 2006; 332:250-1.
3. Farov P, Alali J, Klein MD. Clinical risk factors for gastrochisis and omphalocele in humans: a review of the literature. Pediatr Surg Int. 2010; 26:1135-48.
4. Corey KM, Hornik CP, Laughon MM, McHutchison K, Clark RH, Smith PB. Frequency of anomalies and hospital outcomes in infants with gastrochisis and omphalocele. Early Human Dev. 2014; 90:421-4.
5. Benjamin B, Wilson GN. Anomalies associated with gastrochisis and omphalocele: analysis of 2825 cases from the Texas Birth Defects Registry. J Pediatr Surg. 2014; 49:514-9.
6. Christison-Lagay ER, Kelleher CM, Langer JC. Neonatal abdominal wall defects. Semin Fetal Neonatal Med. 2011; 16:164-72.
7. Arnold MA, Chang DC, Nabaweesi R, Colombani PM, Bathurst MA, Mon KS, et al. Risk stratification of 4344 patients with gastrochisis into simple and complex categories. J Pediatr Surg. 2007; 42:1520-5.
8. Kassa AM, Lilja HE. Predictors of postnatal outcome in neonates with gastrochisis. J Pediatr Surg. 2011; 46:2108-14.
9. Clark RH, Walker MW, Gauderer MW. Factors associated with mortality in neonates with gastrochisis. Eur J Pediatr Surg. 2011; 21:21-4.
10. Van Eijck FC, Hoogeveen YL, van Weel C, Rieu PN, Wijnen RM. Minor and giant omphalocele: long-term outcomes and quality of life. J Pediatr Surg. 2009; 44:1355-9.
11. Marshall J, Salemi JL, Tanner JP, Ramakrishnan R, Feldkamp ML, Marengo LK, et al. Prevalence, correlates, and outcomes of omphalocele in the United States, 1995–2005. Obstet Gynecol. 2015; 126:284-93.
12. Goetzinger KR, Tuulli MG, Longman RE, Huster KM, Odibo AO, Cahill AG. Sonographic predictors of postnatal bowel atresia in fetal gastroschisis. Ultrasound Obstet Gynecol. 2014; 43:420-5.
13. Brantherg A, Blaas HG, Haugen SE, Eik-Nes SH. Characteristics and outcome of 90 cases of fetal omphalocele. Ultrasound Obstet Gynecol. 2005; 26:527-37.
14. Ledbetter DJ. Congenital abdominal wall defects and reconstruction in pediatric surgery: gastrochisis and omphalocele. Surg Clin N Am. 2012; 92:713-27.
15. Bucher BT, Mazotas IG, Warner BW, Saito JM. Effect of time to surgical evaluation on the outcomes of infants with gastrochisis. J Pediatr Surg. 2012; 47:1105-10.
16. Erdoğan D, Anıl MN, Çavuşoğlu YH, Tuncer IS, Karaman I, Karaman A, et al. 11-year experience with gastrochisis: factors affecting mortality and morbidity. Iran J Pediatr. 2012; 22:339.
17. de Buys Roessingh AS, Damphousse A, Ballabeni P, Dubois J, Bouchard S. Predictive factors at birth of the severity of gastrochisis. World J Gastrointest Pathophysiol. 2015; 6:228.
18. Schmieding A, Wittekind B, Salzmann-Manrique E, Schloesser R, Rolle U. Decentralized surgery of abdominal wall defects in Germany. Pediatr Surg Int. 2020; 26:1-10.
19. Payne NR, Pfelegaar K, Assel B, Johnson A, Rich RH. Predicting the outcome of newborns with gastrochisis. J Pediatr Surg. 2009; 44:918-23.
20. Watanabe S, Suzuki T, Hara F, Yasui T, Uga N, Naoe A. Omphalocele and gastrochisis in newborn: over 16 years of experience from a single clinic. J Neonatal Surg. 2017; 6:27.
21. Overcash RT, DeUgarte DA, Stephenson ML, Gutkin RM, Norton ME, Parmar S, et al. University of California Fetal Consortium. Factors associated with gastrochisis outcomes. Obstet Gynecol. 2014; 124:551.
22. Melov SJ, Tsang I, Cohen R, Badawi N, Walker K, Soundappan SS, et al. Complexity of gastrochisis predicts outcome: epidemiology and experience in an Australian tertiary centre. BMC Pregnancy Childbirth. 2018; 18:222.
23. Conner P, Vejde JH, Burgos CM. Accuracy and impact of prenatal diagnosis in infants with omphalocele. Pediatr Surg Int. 2018; 34:629-33.
24. Wright NJ, Langer M, Normen IC, Akkbari M, Wafford QE, Ade-Ajayi N, et al. Improving outcomes for neonates with gastrochisis in low-income and middle-income countries: a systematic review protocol. BMJ Paediatr Open. 2018; 2:e000392.
25. Kumar HR, Jester AL, Ladd AP. Impact of omphalocele size on associated conditions. J Pediatr Surg. 2008; 43:2216-9.
26. Kunz SN, Tiider JS, Whitlock K, Jackson JC, Avasnaino JR. Primary fascial closure versus staged closure with silo in patients with gastrochisis: a meta-analysis. J Pediatr Surg. 2013; 48:845-57.