

Neonatal Outcome of Abdominal Wall Defects at a Tertiary Center in Oman

Abdellatif M^{1*}, Ahmad A¹, Ur Rahman A¹, Al riyami N², Al dughaishi T², Niranjani joshi¹, Zainab Al Balushi¹ and Abdelrahman N¹

¹Department of Child Health, Sultan Qaboos University Hospital, Oman

²Department of Obstetrics and Gynecology, Sultan Qaboos University Hospital, Oman

*Corresponding author: Abdellatif M, Department of Child Health, Senior Consultant Neonatologist and Head of Neonatal Unit, Sultan Qaboos University Hospital, Oman, Tel: 0096897870366; E-mail: mabdelmonem2015@gmail.com

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Abstract

Objective: The aims of this paper are, to evaluate the birth prevalence some of the epidemiological risk factors and neonatal outcomes of newborns with gastroschisis and omphalocele.

Methods: This retrospective descriptive study was conducted between January 2010 and December 2015 at the Sultan Qaboos University Hospital (SQUH) neonatal intensive care unit (NICU).

Results: Ten cases of omphalocele and two cases of gastroschisis were examined. The birth prevalence of gastroschisis and omphalocele was 1.39 in 10000 and 0.28 in 10000 respectively. Antenatal diagnoses were available in six cases (50%). Fifty percent of the cases were inborn. Eight (66.67%) infants were delivered by caesarean section. The median gestational age and birth weight for newborns with gastroschisis at birth were 35 weeks and 2100 g, respectively; for newborns with omphalocele, these values were 37 weeks and 2583 g respectively. The median maternal age for mothers of newborns with gastroschisis was 22.5 y; for mothers of babies with omphalocele, the mean age was 28 y. The median times to full feeding for newborns with gastroschisis and omphalocele were 19 days and 6 days, respectively. The median length of stay in the neonatal unit for newborns with gastroschisis was 35 days; for newborns with omphalocele, the duration was 8.5 days. Fifty percent of all patients exhibited intrauterine growth retardation (IUGR). Primary surgical closure was performed in 10 (83.33%) patients. Associated cardiac anomalies were detected in seven babies (58.33%). Chromosomal anomalies were only documented in two patients with omphalocele. Mortality was documented in three infants (25%).

Conclusion: There were more admissions for patients with omphalocele in comparison with gastroschisis with low birth prevalence compared to reports from western countries. The majority of patients were delivered by cesarean section. Mortality occurred only among patients with omphalocele.

Keywords: Omphalocele; Gastroschisis; Abdominal wall defects

Introduction

The incidence of abdominal-wall defects has been reported to be 1 in 2000 live births. Gastroschisis and omphalocele are the two most common abdominal wall defects. Less common abdominal wall defects include the limb-body wall complex, cloacal and bladder exstrophies, ectopia cordis and urachal cysts [1]. Abdominal-wall defects are usually diagnosed in the first trimester and are characterized by herniation of the abdominal viscera [2]. The estimated birth prevalence of gastroschisis in western countries is twice that of omphalocele. According to European registries, including the EUROCAT network, the incidences of gastroschisis and omphalocele were 3.09 and 3.29 per 10,000 births, respectively, with a live birth prevalence of 2.63 per 10,000 for gastroschisis and 1.13 per 10,000 for omphalocele [3]. Studies from developed countries including the United States, Europe and Japan have indicated that the frequency of gastroschisis is increasing, whereas the birth incidence of omphalocele has remained steady over time [4,5]. The difference in the trends of the two diseases could be linked to the etiologies of both conditions. The etiology of abdominal wall defects is not clear however it has been reported that

gastroschisis is associated with young maternal age, alcohol consumption and smoking while omphalocele is more associated with chromosomal abnormalities and advanced maternal age; the cause of this is not clear. The genetic origin of abdominal wall defects is inadequately understood. The main antenatal diagnostic techniques are ultrasonography and measurement of serum alpha-fetoprotein [6,7]. The majority of abdominal-wall defects are identified by antenatal ultrasound, which has sensitivity ranging from 60% to 75% and a specificity of 95% for both omphalocele and gastroschisis [8].

The incidences in sub-Saharan Africa and the Middle East region have not been determined because no population-based studies of the region have been conducted [3]. Since no studies have been published on the epidemiology of abdominal wall defects in the Middle East, this study was conducted to explain the condition in a tertiary neonatal intensive care unit in Oman.

The aims of this paper are to review our experience of managing newborns with omphalocele and gastroschisis; briefly review the literature; and determine the rate of antenatal diagnosis, sex, gestational age at birth, birth weight, maternal age, mode of delivery, length of hospital stay, time to full feeding, number of babies with intrauterine growth retardation (IUGR), performance of primary or

secondary surgical closure and neonatal complications following surgery and the relevance of associated malformations, chromosomal anomalies and mortality.

Method

Clinical setting

Sultan Qaboos university hospital has an obstetric unit, but also accepts high risk deliveries from other health centers and peripheral hospitals in the region. The newborn service operates as a neonatal intensive care referral unit for pediatric surgery from all over Oman. The neonatal intensive care unit provides intensive care for up to 24 patients.

Study design and data collection

This was a retrospective descriptive study of all newborns with abdominal-wall defects at the Sultan Qaboos University Hospital (SQUH) from January 2010 to December 2015. Data was collected on predesigned and approved data collection forms from the admissions and discharge registers at the NICU.

Statistical analysis was performed using State 12 to determine the means and standard deviations for continuous variables and frequencies for nominal and ordinal variables. The results are expressed as means \pm standard deviations. The study was approved by the local committee of the SQU ethical board (2014/1519/-31/3).

Results

A total of 12 cases of omphalocele and gastroschisis were managed at our center: two (16.67%) of gastroschisis and 10 (83.33%) of omphalocele. Seven of the newborns were males and 5 were females. Fifty percent of the cases were inborn; the other fifty percent were referred from peripheral hospitals. The results of antenatal ultrasound diagnosis were available for only six (50%) patients. The mean gestational age for newborns with gastroschisis at birth was 35 ± 1.41 weeks (range: 34–36 weeks) and the median was 35 weeks; the mean gestational age for newborns with omphalocele was 36.2 ± 3.08 weeks (range: 29–40 weeks) and the median was 37 weeks. The mean birth weight for newborns with gastroschisis was 2100 ± 424.26 g (range: 1800–2400 g) with a median birth weight of 2100 g, and the mean birth weight for patients with omphalocele was 2601.5 ± 463.11 g (range: 1750–3415 g) with a median birth weight of 2583 g.

The mean maternal age for newborns with gastroschisis was 22.5 ± 6.4 y (range: 18–27 y) and the median was 22.5 y; the mean maternal age for mothers of babies with omphalocele was 28.4 ± 5.6 y (range: 21–36 y) and the median was 28 y.

Three babies (25%) were diagnosed antenatally in SQUH by ultrasound. Of the remaining nine patients (75%); only three were diagnosed antenatally in peripheral hospitals; the other six were diagnosed after birth, delivered in peripheral hospitals and transferred after birth to the SQUH neonatal intensive care unit for further management.

Eight babies (67.67%) were delivered by caesarean section, and the remaining four cases (33.33%) were delivered by spontaneous vaginal delivery.

The mean time to full feeding for newborns with gastroschisis was 19 ± 5.66 days (range: 15–23 days), with a median of 19 days, whereas

the mean time to full feeding for newborns with omphalocele was 9.7 ± 9.31 days (range 0–29 days), with a median of 6 days. The average length of stay in the neonatal unit for newborns with gastroschisis was 35 ± 14.14 days (range: 25–45 days), and the median was 35 days; the average length of stay for newborns with omphalocele was 13.6 ± 12.84 days (range: 1–45 days), and the median was 8.5 days.

Six (50%) newborns exhibited IUGR; five (50%) newborns with omphalocele and one with gastroschisis. Neonatal complications following surgery occurred in three newborns: one with omphalocele developed sepsis, one with omphalocele developed post-operative bleeding and died, and one with gastroschisis developed necrotizing enterocolitis. Chromosomal anomalies were documented in two patients with omphalocele: one had trisomy 13, the other had trisomy 18. One newborn with omphalocele was diagnosed with Beckwith-Wiedemann syndrome based on the clinical features. Out of ten babies with omphalocele seven (70%) had congenital heart disease. Three of the 12 babies died, all of whom had omphalocele.

Discussion

To our knowledge this is the first observational study conducted in the Middle East to describe abdominal wall defects. In the present study, all twelve of the infants underwent antenatal ultrasound; however, the diagnosis of abdominal-wall defects was possible in only six (50%) patients. All three patients scheduled at the SQUH were successfully diagnosed by antenatal ultrasound, while only three patients were diagnosed antenatally in peripheral hospitals. The remaining six patients (50%) were diagnosed after birth in peripheral hospitals, which might indicate a lack of ultrasound expertise as the value of an ultrasound is affected by the experience of the radiographer and the timing of the study. Although an increase in the level of maternal serum alpha-fetoproteins is a characteristic of omphalocele and gastroschisis [6,7], this information was not available for our patients.

In this study, the median maternal age for the patients with gastroschisis was lower than that for Omphalocele, the reason for which is not yet clear in the literature [6,7]. A significantly higher number of emergency caesarean deliveries occurred among our patients (66.67%). Few published studies have reported the timing and delivery route of pregnancies with prenatally diagnosed gastroschisis, and the timing and delivery method of such pregnancies remain debatable. Many institutes recommend a planned preterm birth at 36–38 weeks for newborns with gastroschisis; however, studies on this topic have resulted in inconsistent and contradictory reports. Furthermore, most published data report a mean gestational age and spontaneous delivery at approximately 36–37 weeks. This is similar to data reported in a small randomized trial that was included in a recent systematic review of preterm birth for infants with gastroschisis, but the review did not draw a firm conclusion [9]. Data on caesarean sections are inconsistent and do not show any benefit over vaginal delivery. However, the delivery must be performed carefully because trauma to the exposed viscera can occur during either mode of delivery [10]. Moreover, most experts advocate delivery in a tertiary center with neonatal intensive care and pediatric surgery services. Although this would probably lessen the morbidity, there is no firm evidence to date that premature delivery of fetuses with gastroschisis is beneficial for the outcome, however it has been reported that 30% of patients with gastroschisis are delivered prematurely [11]. At our organization, the policy is to deliver infants with gastroschisis by induced vaginal delivery at approximately 37 to 38 weeks because these babies are more likely to

develop IUGR and are more prone to fetal demise. In the management of gastroschisis, the main difficulties are associated with the prevention of late intrauterine death. In the present study, one patient with gastroschisis had IUGR and an emergency caesarean section at 36 weeks owing to fetal distress; this patient was diagnosed antenatally and delivered at the SQUH. The other patient had premature rupture of the membranes and was delivered by spontaneous vaginal delivery at 34 weeks in a peripheral hospital and diagnosed after birth. Likewise the indications for preterm delivery in mothers with fetal omphalocele have not been established, and the mode of delivery for such fetuses is dictated by obstetric indications, as no controlled randomized trials have addressed this issue [12,13]. A caesarean section is the preferred method of many clinicians to deliver fetuses with major defects, to avoid sac rupture or liver damage during delivery [14,15]. However, in a recent study by Kleinrouweler, et al. 17 of 21 (81%) infants were delivered via vaginal delivery and the remaining four by caesarean sections performed entirely on the basis of obstetric indications instead of complications such as liver herniation. Liver herniation was present in 47% of vaginal deliveries with no complications, such as sac rupture and liver hemorrhage [16]. The policy in our institute is that all babies with omphalocele who have been delivered by spontaneous vaginal delivery with the exception of those who are shown to have liver herniation on antenatal ultrasound examinations. The majority of the infants with omphalocele (70%) have been delivered by caesarean section. Three infants were delivered by emergency caesarean section; for one infant delivered at a gestational age of 39 weeks at the SQUH, the indication for caesarean section was fetal distress and potential placental abruption. For the other two infants, the indication was a breech presentation; one was delivered at 35 weeks in a peripheral hospital and the other was delivered at 37 weeks at the SQUH. Four newborns were delivered by elective caesarean section; one mother underwent an elective caesarean section at 37 weeks because she had previously undergone a caesarean section in a peripheral hospital. Two infants who had omphalocele with liver herniation were delivered by elective caesarean section at 36 and 37 weeks at the SQUH. Another infant was delivered by caesarean section at 35 weeks in a peripheral hospital, but the reason was not clear.

Fifty percent of patients in this study had IUGR. Gastroschisis is often associated with IUGR. It is not well understood why those infants develop IUGR, but it may be related to anomalies of the placenta and "direct nutritional wasting secondary to the exposed viscera" [17]. The increase in length of hospital stay and time to full enteral feeding in the two patients with gastroschisis might also be explained by inutero damage to the exposed viscera [8]. In the present study, one newborn with gastroschisis had IUGR, and 50% of the babies with omphalocele had IUGR, although patients with omphalocele are known to have a lower incidence of IUGR. Despite the fact that IUGR is more common in newborns with gastroschisis, chromosomal abnormalities have been reported primarily in newborns with omphalocele; were the estimated prevalence is 30%, and trisomy 13, 18, and 21 are the most common abnormalities [18]. Two infants with omphalocele in this paper had chromosomal abnormalities; one infant with trisomy 13 was delivered by normal spontaneous vaginal delivery in a peripheral hospital, and the other newborn diagnosed antenatally with trisomy 18 was delivered by normal spontaneous vaginal delivery at the SQUH, but died in the hospital. Almost 10% of babies with omphalocele have been reported to have Beckwith-Wiedemann syndrome. One infant with omphalocele was diagnosed with Beckwith-Wiedemann syndrome based on clinical features (organomegally, macroglossia, abdominal-wall defects and gigantism,

neonatal hypoglycemia and single transverse ear crease). Further related abnormalities include CHARGE (coloboma, heart defects, choanal atresia, mental retardation, and genitourinary and ear anomalies) and VACTERL (vertebral, anal, cardiac, tracheoesophageal, renal, and limb deformities) [8]. The prenatal diagnosis of an omphalocele should prompt a careful search for associated anomalies as well as discussion of prenatal invasive testing.

Published data have noted the link between certain anterior abdominal-wall defects, particularly gastroschisis and omphalocele, and congenital heart disease (CHD). Whereas gastroschisis has a weak association with CHD, omphalocele has been reported to have an increased association with CHD [18-20]. In this study, associated cardiac anomalies were detected in seven babies with abdominal-wall defects. Both infants with gastroschisis had atrial septal defects (ASD). Five infants with omphalocele which is forty percent of newborns with omphalocele had associated cardiac anomalies: two had ASD, one had a ventricular septal defect, one had ASD and one had persistent pulmonary hypertension with PDA. It is not yet clear why patients with abdominal-wall defects tend to have a higher incidence of CHD. It has been hypothesized that the embryological development of these patients plays a role in the frequent occurrence of these defects [21].

In the management of gastroschisis, the main difficulties are associated with the prevention of late intrauterine death, and in the management of omphalocele, the main difficulties include the exclusion of other associated conditions and anomalies that were not diagnosed antenatally. The survival outcome in these cases has greatly improved in recent years due to advances in prenatal diagnosis and neonatal anesthetic and surgical techniques. Primary surgical closure is the ideal technique for both gastroschisis and omphalocele. This approach has been reported to reduce the number of days on mechanical ventilation and the length of the hospital stay as well as decrease the rates of infection and mortality. A silo closure can be used in cases of gastroschisis when primary closure is not possible, depending on the degree of viscerobdominal disproportion [22,23]. All patients in this study had primary surgical closure with the exception of two patients with omphalocele with liver herniation who had staged surgical repair to avoid intracompartment syndrome.

Although it is recommended to deliver newborns with abdominal wall defects in a tertiary center the lack of expertise in antenatal ultrasonography make it difficult. The presence of congenital anomalies and the size of the abdominal-wall defect at birth in infants with omphalocele are significant predictors of mortality [17]. Mortality has been reported to be higher among patients with omphalocele, with a rate of 10% as compared to 4-7% among those with gastroschisis [8]. The outcome for newborns with omphalocele is associated with the existence of other concomitant chromosomal and congenital anomalies (most commonly cardiac defects). In the present study, three patients with omphalocele died, two of whom had trisomy 13 and 18, and the third died of sepsis post-surgery. The long-term morbidity in patients with omphalocele is related mainly to respiratory and feeding difficulties associated with a prolonged hospital stay, and 15% of patients develop paralytic ileus, wound infections, and sepsis (8, 22). Indeed, in contrast to patients with omphalocele, the mortality and morbidity in patients with gastroschisis are directly associated with the severity of the gastrointestinal disease, and the majority of those deaths are related to extensive necrosis, short-bowel syndrome and multiple atresia [8,24]. This might be explained by the fact that the size of the abdominal-wall defect and duration of exposure to amniotic fluid are factors in adverse long-term effects on gut motility and function in

patients with gastroschisis [22]. Our study reflected that the average length of hospital stay and the time to full feeding were increased in both patients with gastroschisis compared with those in patients with omphalocele. Only one patient with gastroschisis developed complicated bowel pathology with necrotizing enterocolitis.

Previous reports from Oman revealed that congenital anomalies are a major cause of death and disability in the country [25-27]. This could be explained partially by the high degree of consanguineous marriages in the country. Although the prevalence of gastroschisis in the west seems to be increasing, the majority of the patients admitted to our unit had omphalocele. This difference might be linked to genetic and environmental reasons as the sketch and incidence of congenital malformation may fluctuate over time or geographical location, thus reflecting a complex interaction of known and unknown genetic and environmental factors including sociocultural, racial and ethnic variables [28].

Since the average number of live births in Oman is 72,109/yr during the study period from 2010 to 2015 the incidence of omphalocele will be 1.39 in 10,000 and 0.28 in 10,000 for gastroschisis which is much smaller compared to figures from the western world. However,

Despite the high incidence of consanguinity in the country the birth prevalence of abdominal wall defects is low compared to other western countries. The lack of reliable published data on the pattern of congenital malformation in the country makes it difficult to calculate the exact birth prevalence and incidence.

The results of the present study are limited by its retrospective nature and the small number of patients in both groups, as well as the fact that information on some patients with abdominal wall defects who were still born and those who died before admission to the NICU was not available. This may influence the results of the study.

Conclusion

We observe that the most common abdominal-wall defects observed in our unit are omphalocele, most likely due to genetic reasons related to high consanguinity in the country; however the number of patients in this study is small to draw a solid epidemiological conclusion.

The number of cesarean sections is noted to be high. Early detection of those patients in peripheral hospitals and referral to a tertiary center for planned delivery may reduce the number of cesarean sections. Further research might be required to determine the optimum mode of delivery in patients with gastroschisis and omphalocele and the role of cesarean section and/or preterm delivery in fetuses with gastroschisis

What is already known in this topic

Gastroschisis and Omphalocele are the two commonest types of abdominal wall defects admitted to the neonatal intensive care unit. Recent reports from western countries revealed that the incidence of gastroschisis is on the rise yet there are no reports from the sub-Saharan Africa and the Middle East region. The optimum mode of delivery is not yet clear.

What this study adds: Our results from this observational study show that, unlike reports from the west, we had lower birth prevalence for patients with gastroschisis in Oman as compared to omphalocele. Further epidemiological studies and establishment of a database are warranted for congenital malformations from Oman and the Middle

East, to determine the incidence and birth prevalence since consanguineous marriage is significant in this part of the world. Lack of antenatal detection and centralization of delivery might have contributed to the high rate of cesarean sections in patients with abdominal wall defects at our institute.

Author Contribution

Conception and design: Mo and Na; Acquisition, analysis and interpretation of data: Mo, Na, Ni; Drafting the article: Jo and Ta; Revising it critically for important intellectual content: As and As; Approved final version of the manuscript: Mo and Ze.

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