Original Article

Changing Trend in the Management of Omphalocoele in a Tertiary Hospital of a Middle-Income Country

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Abstract

Background: The peri-operative management of omphalocoele in low- and middle-income countries is challenging owing to non-availability of neonatal intensive care units and equipment needed for the care of this anomaly. **Aim:** This study examined our experience in the management of omphalocoele and compared the pattern and outcome with a similar study from the same centre conducted four decades ago. **Methods:** A retrospective study of neonates managed for omphalocoele from 2003 to 2017 (Group A) was performed. Their demographic characteristics, clinical presentation, management modality and outcome were obtained and statistical analysis was performed to determine the predictors of mortality. The findings were also compared with the findings of a similar study (Group B) published from this centre four decades ago from 1973 to 1978. **Results:** A total of 95 patients were managed in Group A and 33 in Group B. Their ages ranged from 1 to 15 days with a median age of 1 day and a median gestational age at birth of 37 weeks (range – 36–43 weeks) in Group A and 5–72 h in Group B. There were 54 (56.8%) boys and 41 (43.2%) girls in Group A and 17 (51.5%) boys and 16 (48.5%) girls in Group B. Rupture of the sac was observed in 18 (18.9%) patients in Group A and 13 (39.4%) in Group B. Operative management was adopted for 55 (57.9%) patients in Group A compared to 14 (42.4%) in Group B. Mortality was recorded in 16 (16.8%) patients in Group A and 16 (48.5%) in Group B. Following further analysis in Group A, management outcome was noted to be significantly associated with the state of the sac ($P = 0.011$), presence of associated sepsis ($P = 0.002$) at presentation and management modality ($P = 0.048$) with only associated sepsis independently predicting mortality. **Conclusion:** Although epidemiological trend and clinical presentation are still similar, management outcome has improved over the years.

Keywords: Changing trends, management, middle-income country, mortality, omphalocoele, outcome, sepsis

Introduction

The appearance of anterior abdominal wall defects in the newborn is a source of psychological disturbance to the mother and this is particularly worse in a neonate with a major omphalocoele in which many of the abdominal viscera are visible through the covering membrane.[1] Although the prevalence of omphalocoele in Africans is not known, it varies from 0.9 to 3.8 per 1000 births in Europe and America.[2] There, however, appears to be an increase in the prevalence of omphalocoele in Asia, especially in Japan and Singapore.[3,4] In spite of the grotesque appearance of the anterior abdominal wall in children with omphalocoele, they present late in most low- and middle-income countries (LMIC) when sepsis would have set in with consequent deterioration in their clinical state.[5,6] Thus, the management of omphalocoele is challenging, especially in these countries, and this is often as a result of the reduced capacity of the abdomen which makes closure of the large defects to be difficult, presence of associated congenital malformations[5,7-9] and lack of appropriate equipment with no dedicated neonatal intensive care unit (NICU) for post-operative care.[10] Conversely, the management outcome has improved in high-income countries (HIC)[11] while still poor in LMIC[8,9,12] with overall mortality ranging from 30% to 45%. In view of some of the challenges (late presentation, lack of appropriate equipment and lack of dedicated NICU) and outcome highlighted above, this study presents our experience in the management of

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omphalocoele and compares the pattern and outcome with a similar study from the same centre conducted four decades ago.

Methods

All neonates managed for omphalocoele over a 15-year period from January 2003 to December 2017 were retrospectively studied. Information about the demographic characteristics of the neonates, clinical presentation, the state of the covering sac (whether it was intact or ruptured), mode of management and outcome were extracted from the clinical records of these patients (Group A) and entered into a pro forma. Data from a publication on the second cohort of patients (Group B) managed for omphalocoele in the same hospital four decades ago from 1973 to 1978[13] were obtained for comparison. Categorical variables were presented using frequency, proportions and ratios, while continuous variables were summarised using mean and standard deviation or median and interquartile range depending on their distribution. Chi-square and Fisher's exact test analyses were used to test the association between categorical variables and mortality, while independent sample *t*-test or Mann–Whitney U test was used to compare continuous variables depending on their distribution. Multivariate logistic regression analysis was then performed to determine which of the statistically significant associated factors independently predicted mortality. Analysis of the data obtained was performed using Statistical Package for the Social Sciences (SPSS Inc., Chicago, IL. USA) Version 23.0 Software with the level of significance set at $P < 0.05$. Ethical approval for the study was obtained from the University of Ibadan/University College Hospital Ethical Review Committee.

Results

A total of 95 neonates were managed during this period, their ages ranged from 1 to 15 days with a median age of 1 day and a median gestational age at birth of 37 weeks (range – 36–43 weeks). The mean age of their mothers was 27.9 ± 3.6 years. A total of 67 (70.5%) neonates were presented within the first 3 days of life. Of these, 44 (46.3%) neonates were presented within 24 h of life. The mean birth weight was 3.0 ± 0.5 kg, with a range of 2.2–4.1 kg, and the mean weight at presentation was 3.1 ± 0.5 kg. There were 54 (56.8%) boys and 41 (43.2%) girls (male-to-female ratio = 1.3:1). Only 10 (10.5%) patients had prenatal diagnosis of anterior abdominal wall defect and this was observed in patients who had antenatal care in our hospital. The diameter of the defects was >4 cm in 56 (59%) neonates and the mean diameter of the defects was 6.7 ± 3.0 cm with a range of 3–14 cm. Co-morbidities were observed in 21 (22.1%) patients, these comprised neonatal sepsis in 7 (7.4%), anaemia in 4 (4.2%), jaundice in 4 (4.2%), retroviral exposure in 2 (2.2%) and perinatal asphyxia and inguinal hernia in 1 (1.1%) neonate each. The covering sac was intact in 77 (81.1%) patients and ruptured in 18 (18.9%) patients. Associated congenital anomalies were observed in 14 (14.7%) which included Beckwith–Wiedemann Syndrome in 5 (5.3%) patients and polydactyly in 2 (2.1%) patients [Table 1]. Surgery was performed on 55 (57.9%) patients, 39 (41.0%) patients had non-operative management and only 1 (1.1%) patient refused any form of management and subsequently was discharged. Of the patients who had surgical management, 53 (55.8%) had primary closure of the defects (which include 39 [41%] patients whose defects were 4 cm and below and 14 [14.7%] whose defects were 4 cm–6 cm in diameter irrespective of the state of the covering membranes), whereas 2 (2.1%) had silo constructed to temporarily house the herniated viscera.

Post-operative complications were reported in 14 (14.7%) patients and these comprised wound-related complications in 12 (12.6%) patients (surgical site infection in 10 (10.5%) and burst abdomen and flap necrosis in 1 (1.1%) patient each) and haemorrhage and adhesive bowel obstruction in 1 (1.1%) patient each. Mortality was recorded in 16 (16.8%) patients; these included 9 (9.5%) patients with ruptured sac and 7 (7.4%) patients with intact sac, 2 (2.1%) patients with defects <4 cm and 14 (14.7%) patients with defects >4 cm ($P = 0.707$). The median length of hospital stay among the survivors was 18 days, with a range of 4–90 days. Patients with major omphalocoele were managed non-operatively with regular dressings using saline and 1% silver sulphadiazine cream and were discharged once the wound appeared clean, contracted and appeared manageable on an outpatient basis. Univariate analysis [Table 2] revealed that the management outcome was significantly associated with the state of the covering sac, whether intact or ruptured ($P = 0.011$), the presence of associated sepsis ($P = 0.002$) at presentation and the modality of management ($P = 0.048$). Following multivariate analysis, only associated sepsis independently predicted mortality with more than five times as much risk of death [Table 3]. Mortality in patients with ruptured omphalocoele was four-and-a-half times as likely as those with intact membrane, but this was not statistically significant ($P = 0.070$).

A total of 33 patients were managed within the 5 years in Group B. The age range of the patients was 5–72 h and the male-to-female ratio was 1:1. Rupture of the covering membrane was observed in 13 (39.4%) patients in Group B in comparison with 18 (19.0%) in Group A and 16 (48.5%)

Table 1: Associated congenital anomalies

Congenital anomalies	Frequency (%)
BeckwithWiedemann Syndrome	5 (35.72)
Polydactyly	2 (14.30)
Malrotation of the intestine	1 (7.14)
Intestinal atresia	1 (7.14)
Patent urachus	1 (7.14)
Undescended testis	1 (7.14)
Hydrocoele	1 (7.14)
Cleft lip and palate	1 (7.14)
Lower midline association	1 (7.14)
Total	14 (100.00)

Table 2: Factors associated with mortality

	Alive, *n* (%)	Dead, *n* (%)	*P*
Sex			
Male	46 (85.2)	8 (14.8)	0.545
Female	33 (80.5)	8 (19.5)	
Age in years, median (IQR)	1 (2)	1 (2)	0.816
Birth weight (kg)	3.0±0.9	3.1±0.4	0.289
Diameter of defect (*n*=55)			
<4 cm	15 (88.2)	2 (11.8)	0.707
>4 cm	30 (78.9)	8 (21.1)	
State of sac			
Intact	68 (88.3)	9 (11.7)	0.011*
Ruptured	11 (61.1)	7 (38.9)	
Associated sepsis (*n*=88)			
Yes	23 (67.6)	11 (32.4)	0.002*
No	50 (92.6)	4 (7.4)	
Source of referral (*n*=61)			
Rural	8 (66.7)	4 (33.3)	0.263
Urban	40 (81.6)	9 (18.4)	
Management type (*n*=78)			
Non-operative	16 (69.6)	7 (30.4)	0.048*
Operative	49 (89.1)	6 (10.9)	
Mean maternal age (years) (SD)	27±3.5	32.0±3.0	0.050

*Statistically significant at *P*<0.05. IQR: Interquartile range, SD: Standard deviation

Table 3: Multivariate logistic regression analysis of factors predicting mortality in omphalocoele

Variable	β	Adjusted OR	95% CI	*P*
Associated sepsis				
Yes	1.70	5.47	1.01–29.55	0.048*
No				
State of membrane				
Ruptured membrane	1.51	4.52	0.88–23.20	0.070
Intact				
Type of management				
Non-operative	1.44	4.21	0.91–19.57	0.066
Operative				

*Statistically significant at *P*<0.05. OR: Odd ratio, CI: Confidence Interval

patients in Group B had associated anomalies, whereas 14 (14.7%) had in Group A. Non-operative management was used to manage omphalocoele on 19 (57.6%) patients in Group B and 39 (41.1%) in Group A; of these, non-operative management was adopted on all but 1 patient with intact covering membrane in Group B and 33 (34.7%) patients with intact membrane in Group A. Mortality was 48.5% in Group B and 16.8% in Group A [Table 4].

Discussion

The management of omphalocoele is challenging in LMIC. Indeed, it is daunting when the covering membrane is already ruptured before presentation with consequent high morbidity and mortality.[4,5] This may be worsened by late presentation as observed in this study, in which 29.5% of the patients were presented beyond the 3rd day of life up to 15 days. Early presentation and prevention of sepsis are key important aspects of successful management of patients with omphalocoele. The fact that many patients are often delivered at home[12] following an unsupervised antenatal period and have to be transported to the hospital with dirty clothing that tends to contaminate the covering membrane or eviscerated organs makes the prognosis of their management to be poorer in the developing countries.[12,14-18] This is worsened by the lack of requisite facilities to support their perioperative management with consequent high morbidity and mortality rates as is the case in this country.[10] Although there are variations in the prevalence of omphalocoele by maternal age at delivery,[19-21] the mean maternal age in this study appears to be lower than in previous reports that stated that it is more common in mothers older than 40 years.[22,23]

In HIC, prenatal diagnosis, especially of major omphalocoele,[24,25] *in utero* transfer to the nearest paediatric surgical centre, assisted delivery to prevent rupture and contamination of the covering membrane and availability of NICU and other requisite facilities needed for primary peri-operative management of these patients have made the management outcomes to be very good.[16,26-29] It has been reported that only about two-third of cases of anterior abdominal wall defects are detected with prenatal ultrasound. Furthermore, the rate of false-positive reports and misdiagnoses of anterior abdominal wall defects is very high. These have been attributed to the technique of scanning, the quality of equipment and the experience of the sonologist.[30,31] The observed low rate of prenatal diagnosis of 10.5% in the present study compared to previous reports[30,31] may be attributed to the use of ultrasonographic machines with very poor resolution and the fact that majority of the sonologists might have been poorly trained in the use of ultrasonography in the peripheral centres.[30,31] This may also explain why the rate of prenatal diagnosis was lower than the reported rate from HIC,[30,31] although it is higher than the reported rate from another centre in this country.[8]

The size of the defect is a very strong factor to be considered in LMIC due to the presence of associated severe congenital anomalies, lack of appropriate equipment, lack of dedicated NICU for post-operative care and inadequate provision of safe anaesthesia.[10,32,33] Improvements in neonatal ventilation, peri-operative monitoring, nutrition and availability of synthetic materials like soft mesh[34-37] to achieve primary closure of omphalocoele with very large defects have also reduced the peri-operative morbidity and mortality in HIC.[27] Non-operative management of major omphalocoele with intact membranes or severe associated congenital malformations is still being practiced in LMICs, whereas primary fascial closure of small defects, staged closure using silos and conversion to incisional hernia (by closing the skin only over the defect) are adopted for the large defects and those with ruptured membranes.[16]

Table 4: Comparison of present study with previous study from the same centre

Parameters	Group A (current study)	Group B (Nwabueze-Ihekwaba[13])
Number of cases	95	33
Study period (years)	15	5
Age range	1–15 days	5–72 h
Sex ratio (male:female)	1.3:1	1:1
Type of exomphalos, *n* (%)		
Intact membrane	77 (81.1)	20 (60.6)
Ruptured membrane	18 (19.0)	13 (39.4)
Associated anomalies	14 (14.7)	16 (48.5)
Treatment, *n* (%)		
Non-operative	39 (41.1)	19 (57.6)
Operative	55 (57.9)	14 (42.4)
Mortality	16 (16.8)	16 (48.5)

Expectedly, mortality was high in patients with ruptured sac (38.9%). In the present study, the management outcome was significantly related to whether the covering sac is ruptured or not, associated sepsis[9,13,16,38] and the management modality adopted but only the presence of associated sepsis independently predicted mortality as suggested by multivariate analysis.

A comparison of the findings in this study (Group A) with an earlier study carried out about four decades ago in this centre revealed a similar trend in the epidemiology and presentation of the anomaly. However, the number of patients with ruptured omphalocoele was high in Group B in comparison to Group A. The fact that obstetric scan for prenatal diagnosis was not available in this environment four decades ago[39] and an accompanying traumatic vaginal delivery of patients with large defects[40,41] may account for this. Furthermore, recent improvements in perinatal services occasioned by proper training of traditional birth attendants to recognise high-risk pregnancies that can lead to traumatic deliveries and consequent rupture of the covering sac may explain the low incidence of ruptured omphalocoele in Group A patients.[42] However, there is a change in the trend of management of omphalocoele as operative management was employed more on Group A patients than Group B patients. Operative management was adopted to manage all the ruptured cases and one patient with an intact membrane in Group B, whereas in Group A, operative management was adopted for all the patients except those with very large defects and associated severe congenital anomalies. Four decades ago, the specialty of paediatric surgery in the country was just being established and non-operative management was more favoured because of lack of trained paediatric anaesthetists, non-availability of NICU and lack of requisite equipment for peri-operative management of these neonates.[10,32] These factors may also explain the reduction in the mortality rate observed in Group A patients.

The retrospective nature of this study poses a substantial limitation as the data were retrospectively collected. The data may leave out some variables due to inaccurate recording and mistakes in retrieving the records.

Conclusion

The epidemiology and presentation of omphalocoele are still the same; however, there is a change in the trend of management with a gradual reduction in mortality rate with consequent improvement in the management outcome in our centre. The management outcome was significantly related to the state of the covering sac (whether it was intact or ruptured), associated sepsis and the management modality adopted but only the presence of associated sepsis independently predicted mortality. It is therefore recommended that further reduction in mortality rate may be achieved with early presentation and provision of appropriate equipment that can support the perioperative management of these neonates, especially the ones with ruptured omphalocoele.

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Conflicts of interest

There are no conflicts of interest.

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