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Original article

Congenital abdominal wall defect in neonates: difficulty of management and prognosis in developing country

Malformations de la paroi abdominale antérieure : difficultés thérapeutiques et pronostic dans un pays en voie de développement

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Résumé

Introduction : la prise en charge du laparoschisis et de l'omphalocèle n'est pas consensuelle et reste un défi pour les chirurgiens pédiatres des pays en développement avec un taux de mortalité élevé. Devant cette mortalité élevée retrouvée dans de précédentes études réalisées dans notre service, le traitement conservateur était effectué de façon systématique en cas d'omphalocèle non rompue. Cependant, le traitement des omphalocèles rompues et des laparoschisis reste un défi majeur dans notre pratique quotidienne mais également pour les chirurgiens pédiatres africains du fait d'un plateau technique défaillant. Le but de cette étude était de rapporter notre expérience sur la prise en charge et l'évolution de ces malformations en insistant sur les difficultés thérapeutiques.

Méthodologie : nous avons réalisé une étude rétrospective sur 20 ans au centre hospitalier et universitaire de Yopougon (CHU, Abidjan), incluant tous les nouveau-nés traités dans le service pour une omphalocèle ou un laparoschisis. Cette étude a inclus 129 omphalocèles et 30 laparoschisis.

Résultats : le diagnostic prénatal a été fait dans sept cas (4,5 %). L'accouchement s'est fait en dehors d'un CHU dans 143 (90 %) cas. Quatre-vingt-dix pour cent des nouveau-nés ont été transférés en taxi ou en bus sans incubateurs ni perfusions. Le traitement conservateur consistant en l'application d'une solution aqueuse disodique à 2 % a été réalisé pour l'omphalocèle non rompue. Pour les laparoschisis et les omphalocèles rompues, le délai moyen d'intervention était de 28,8 ± 2,64 heures. Dans l'ensemble, 66 (41,5%) bébés sont décédés. La principale cause de décès était l'infection 47 (71 %).

Conclusion : les malformations congénitales de la paroi abdominale antérieure ont un diagnostic anté natal faible avec un taux de mortalité plus élevé dans notre pratique. L'augmentation du taux de survie nécessiterait une amélioration du plateau technique, des soins néonataux adéquats et une assurance maladie.

Mots-clés : Laparoschisis - Mortalité - Nouveau-né - Omphalocèle.

Abstract

Introduction: management of gastroschisis and omphalocele is not consensual and remains challenging for surgeons in developing countries with a higher mortality rate. Faced to hight mortalitity in previous studies in our center, priority was given to conservative treatment in case of unruptured omphalocele. Management of ruptured omphalocele and gastroschisis remain a major challenge in our daily practice but also for African pediatric surgeons due to deficient hospitalcare. The aim of this study was to report the outcome of these CAWDs with highlight therapeutic difficulties.

Methodology: we carried out a retrospective study at Yopougon (Abidjan) teaching hospital, included all newborns with as either gastroschisis or omphalocele managed in our unit from twenty years.This study included 129 omphalocele and 30 gastroschisis.

Results: prenatal diagnosis was done in seven (4.5%) cases. The delivery was done outside of tertiary centre in 143 (90%) cases. Ninety percent of neonates were transferred either by taxis or bus without incubators and intravenous fluids. Conservative treatment with dissodic 2% aqueous was applied for unruptured omphalocele. For gastroschisis and ruptured omphalocele, the mean time for surgery was 28.8±2.64 hours. Overall, 66 (41.5%) babies died. The main cause of death were infections 47 (71%).

Conclusion: congenital abdominal wall defect have marginal prenatal diagnosis with a higher mortality rate in our practice. Increasing the survival rate required improvement of infrastructure, skilled neonate care, availability of prosthetic materials and health insurance.

Keywords: Gastroschisis - Neonate - Mortality – Omphalocele.

Introduction

Gastroschisis and omphalocele also called ″mean cœlosomy" are the two most common congenital anterior abdominal wall defects (CAWD) in neonate [1,2]. There were encountered worldwide with an estimated incidence of 1:4000 live-births for omphalocele and 4 to 5:10000 live births for gastroschisis in Western countries [3,4]. Their management is not consensual [5] and remains a major source of morbidity and mortality in neonate population in developing countries. In recent decades, improvement in diagnostic facilities, development in neonatal intensive care, advances in pediatric anesthesia and resuscitation have improved the survival rate in developed countries from 10% in 1960 to about 90-95% in 1980-1990 [6]. However, in Sub Saharan Africa, management of these CAWDs remains a challenge with a high mortality rate ranging from 30% to 100% [7-9]. In previous study done in our unit, surgical procedure led to 67% mortality for omphalocele [10]. Faced to this situation, priority was given to conservative treatment in case of unruptured omphalocele independently of the defect's size. Management of ruptured omphalocele and gastroschisis remain a major challenge in our daily practice but also for African pediatric surgeons due to deficient hospital care in several studies [8,11,12]. In view of the above, what would be the determining factors of this still high morbidity and mortality of CAWDs in our practice? The aim of this study was to report the outcome of these CAWDs and to highlight therapeutic difficulties.

Methodology

This retrospective study was carried out at Yopougon teaching hospital (Abidjan, Côte d'Ivoire). The study population included all newborns (≤ 28 days of age) with gastroschisis or omphalocele managed in the pediatric surgery unit from January 1998 to December 2017. We excluded newborn that was diagnosed with superior or inferior cœlosomy, prune belly syndrome, those who were transferred to another hospital and those with missing data. Of a total of 224 CAWDs, this study included 159 CAWDs (129 omphalocele and 30 gastroschisis). The mean age of mothers was 26,9 ±5,65 years. No history of exposure to X-rays

irradiation, family history of abdominal wall defects or any other congenital malformations was noted. Maternal demographics data were summarized in table I.

None of babies was transported in incubators, with intravenous fluid or with nasogastric tube. Our patients are usually admitted onto the unit, we do not have neonatal intensive care unit (NICU). All our patients received intravenous broad spectrum antibiotics (ceftriaxone® 100 mg/kg/day and metronidazole® 30mg/kg/day) throughout the period of their admission. Conservative treatment for unruptured omphalocele consisted of a twice a day application of the 2% disodium aqueous eosin on the entire omphalocele translucent membrane independently of the defect's size. Omphalocele sac traction was performed in the early days to prevent downfall and maceration in contact with the skin. This application was continued until the drying and complete epithelialization of the sac. When membrane infection of the sac occured, it was treated with topical treatment with sodium hypochlorite concentrated solution until its complete disinfection. After local disinfection, the twice a day application of dissodic 2% aqueous eosin was continued (figure1).

The new-born was exited, when the mother had a good knowledge and good practice in the application of the dissodic 2% aqueous eosin and the omphalocele had an early epithelialization without infection, vital signs stable, regular bowel movements, normal diet and taking regular weight. Conservative treatment was continued at home by the mother with external follow-up. Ruptured omphalocele and gastroschisis (figure 2) underwent surgery.

Gradual reduction has been managed with an improvised silo under general anesthesia at operative theatre. The silo is fashioned from a sterile urine bag or sterile gloves. They were on partial parenteral nutrition with intravenous 10% glucose containing electrolytes and amino acids (0,15mg/ kg). Postoperatively, IV fluids and continuous nasogastric tube drainage were done. When babies passing stools, we start fractional feeding on breast milk exclusively with monitoring of intestinal transit. The studied variables were prenatal diagnosis, type of management, duration of hospitalization, complications and mortality. The statistical analysis was descriptive and comparative. Comparison of mortality in our series with those of other studies was made with the test of the Khi square test with a significance level of 5% ($p< 0.05$).

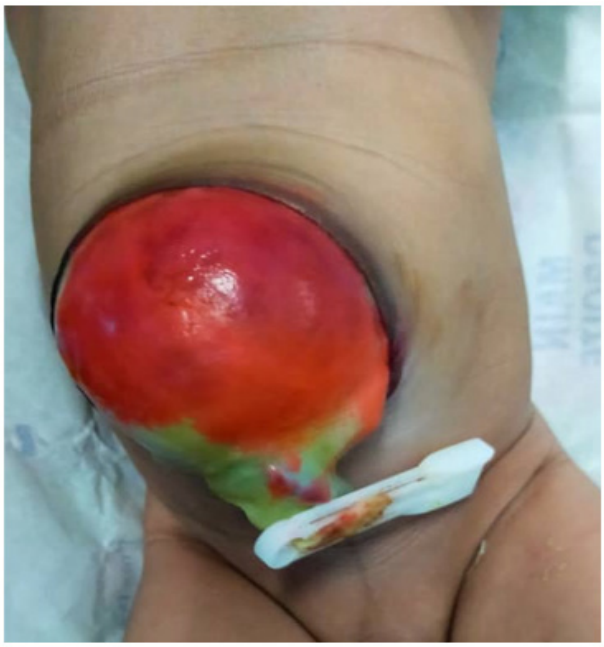

Figure 1: application of the dissodic 2% aqueous eosin on the omphalocele sac soiled by meconium amniotic fluid.

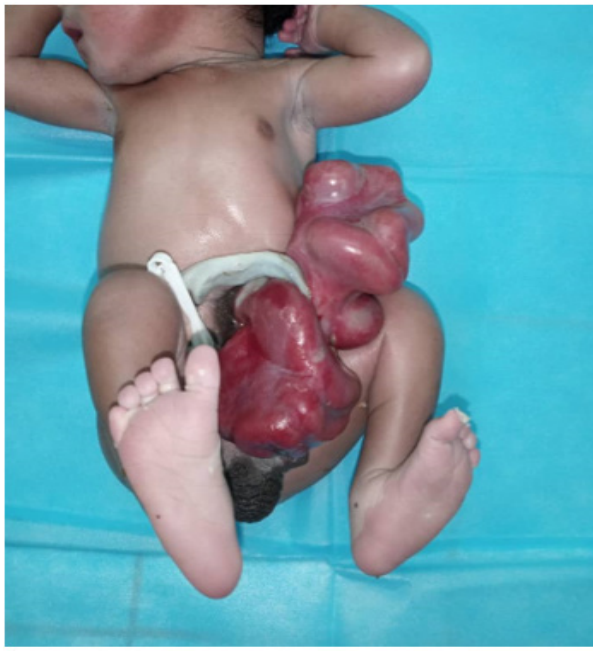

Figure 2: gastroschisis with exposed edematous viscera

Table I: Demographic data of mothers

	Omphalocele	Gastroschisis	Total
Mean age	**28.5 ± 6.52**	**25.3 ± 4.79**	
< 20 years	5	5	10
21 – 30 years	84	24	108
> 30 years	40	1	41
Profession			
Housewife	60	20	80
Student	6	3	9
Official	5	2	7
Tertiary activities	9	5	14
Gestity			
Primigravida	19	12	31
Multigravida	110	18	128
Parity			
Primiparity	45	12	57
Multiparity	84	18	102
Alcohol	2	1	3
Smoking	1	0	1
Consanguinity	4	0	4
Provenance			
Abidjan	43	8	51
Others cities	21	13	35
Village	39	7	46
Not precised	26	2	28
Number of prenatal visits			
0	24	0	24
1 - 3	153	33	186
≥ 4	38	12	50
Number of prenatal ultrasound			
0	45	8	53
1-2	26	11	37
≥ 3	60	12	72

Results

Prenatal diagnosis of CAWDs was done in seven cases (4.5%). Four out of seven fetuses detected prenatally with CAWDs were delivered by caesarean section, and two of them had ruptured omphalocele. The delivery was done outside of teaching hospital in 145 (91.2%) cases; amongst them 19 (12%) occured at home. Babies transportations were not medicalized in 99(62%). The eviscerated organs and omphalocele bag were wrapped with gaze and loinclothes. Overall, 11 (8.5%) patients had ruptured omphalocele. Nine patients (7%) had ruptured omphalocele at birth whereas two (1.6%) had secondary rupture of omphalocele bag. (table II).

For gastroschisis and ruptured omphalocele, the mean time for surgery was 28.8±2.64 hours. The average hospital stay was 13±4.52 days for omphalocele and 7 ±4.74 days for gastroschisis. The average duration for the complete epithelialization was 72±8.2 days

(figure 3).

Mortality was 31% for omphalocele (100% for ruptured omphalocele and 24% for unruptured omphalocele). Mortality was 86% for gastroschisis. The two mains cause of death were infections 47(71%), and anaemia 22 (33%) (table III).

Table II: Demographic and clinical data of newborns

	Omphalocele	Gastroschisis	Total
Prenatal diagnosis	**7**	**0**	**7**
Gestational age			
Preterm	4	8	12
Full term	125	22	147
Place of delivery			
At home	15	4	19
Primary health care	59	16	75
Secondary center	40	9	49
Teaching hospital	12	4	16
Babies transportations			
Medical	47	13	60
Not medical	82	17	99
Bowels protection before admission			
Yes	51	20	71
No	78	10	88
Associated abnormalities			
Heart anomalies	8	0	8
Musculoskeletal	7	0	7
Beckwith Wiedemann syndrome	13	0	13
Gastrointestinal tract	5	0	7
Glycemia			
Hypoglycemia	17	1	18

Table III: treatment outcomes of CAWDs

	Omphalocele		Gastroschisis	Total n (%)
	Not ruptured	Ruptured		
Mean hospital stay (days)	13		5	-
Mean mortality	40 (31%)		26 (86%)	66 (41.5%)
Mortality	29 (24%)	11 (100%)	26 (86%)	66 (41.5%)
Death period				
Preoperative	0	5 (45.5%)	24 (80%)	29 (18.2%)
Peroperative	0	1 (9%)	0	1 (0.6%)
Postoperative	0	5 (45.5%)	2 (6.6%)	7 (4%)
Complications*				
Distress respiratory	10		6	16 (10%)
Bowel occlusion	8		4	12 (7,5%)
Hypoglycemia	11		0	11 (7%)
False route	1		0	1
Dehydration	1		0	1
Infection	27		20	47 (29.5%)
Anemia	6		16	22 (13.8%)

*most neonate have more than one complication

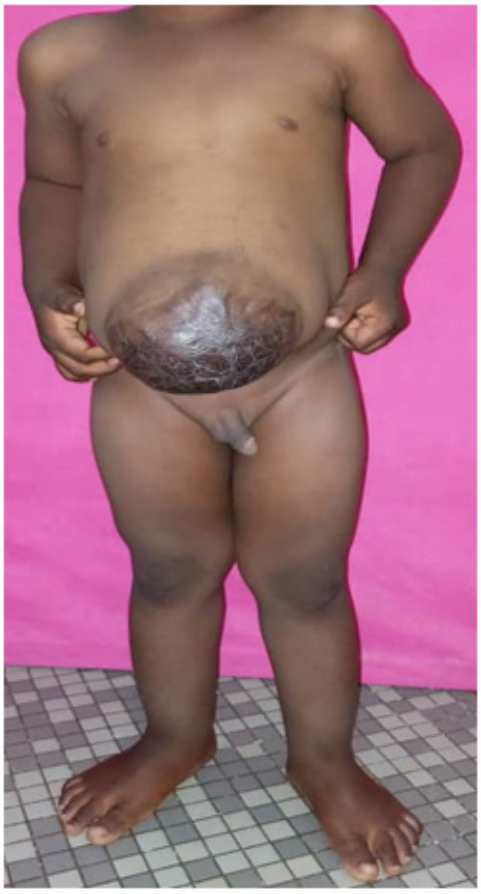

Figure 3: Ventral hernia formed following non-operative management of omphalocele

Discussion

CAWDs are typically diagnosed prenatally using fetal ultrasonography, and affected neonates are treated at a center with access to high-risk obstetric services, neonatology, and pediatric surgery. In developed countries, both were detected prenatally in up to 60 - 95% of cases using obstetric sonography and α-fetoprotein concentrations in maternal blood and amniotic fluid [8,9,13]. The prenatal diagnosis in this series remains low compared with previously reported in western data. This low rate of prenatal diagnosis is common in developing countries: 4% at Bangladesh [14], 2% at Ouganda [7], 0 % at Kano (Nigeria) [8]. This low rate was multifactorial: (a) poor pregnancy monitoring due to low socioeconomic level of our populations, (b) the fact that for majority of the sonologists, congenital malformation detection are not systematic, (c) lack of radiologist in all hospitals. Prenatal diagnosis creates a potential opportunity to counsel the parents and to discuss options for perinatal

management, depending on the type and severity of the defect and the nature of co-morbidity, if any.

Most authors and clinicians encourage delivery at a tertiary center with immediate neonatal and pediatric surgery access [15]. Delayed presentation to tertiary pediatric surgery center is a major problem in the management of CAWDs in low resource settings [1,8,16]. In the current study, most babies were delivered at home or at peripheral health center where the unskilled environment does not led to appropriate provision of initial care for a neonate with a surgical emergency. Then, they were transferred in inadequate conditions, without incubator and initial resuscitative care, and they arrive in the hospital sometimes dehydrated, hypothermic and septic. The unequal distribution of pediatric surgeons in our context is also source of delayed care. Despite the fact that majority (78%) of our neonates were born in hospitals, immediate management of these malformations could not be done on site due to absence of pediatric surgeons. Only three out of 40 (7%) Ivorian pediatric surgeons are in the center of the country; the remote 37 are concentrated in the capital (Abidjan). However, even admitted to tertiary center in Abidjan, care is not optimal due to lack of NICU, insufficiency in trained staff in neonatal resuscitation and they are hospitalized without incubators.

The optimal therapeutic strategy for CAWDs remains undetermined and included three main strategies: primary closure, staged repair and delayed closure [17]. The definitive goal was to reduce the viscera safely and, to close the abdominal wall defect with an acceptable cosmetic appearance. Delaying a delicate surgical and anesthetic procedure in a newborn baby is particularly advantageous in our environment where neonatal anesthesia, parenteral nutrition and NICU are unavailable. Another advantage of a delay in operative repair is that it permits time for thoracic and abdominal growth, which may limit respiratory complications associated with the abdominal wall reconstruction and enable a more successful repair. The disadvantages were infection and the lost of view of patients after epithelialization obtained at a mean 70 days. However, gastroschisis and ruptured omphalocele mandates immediate operative intervention which was a high challenge for us. Primary closure or staged repair of these conditions without prosthetic meshes, preformed silos, parenteral nutrition and mechanical ventilators exposed to 86-100% mortality in our practice. Ameh et al [18], reported a mortality rate of 60% for ruptured omphalocele and gastroschisis in Nigeria, while several authors have found a mortality rate up to 90% for gastroschisis [12,19]. This high mortality in Sub Saharan Africa is not surprising considering the myriad problems faced by our patients including lack of prenatal diagnosis, late presentation, poor means of transportation, financial constraints, significant infrastructure gaps, lack of a neonatal intensive care and total parenteral nutrition, poor monitoring, insufficiency in trained staff in neonatal resuscitation and surgical equipment, dearth of trained support personnel, lack of proper prosthetic covering material [9]. In developed countries, recent published studies reported mortality rate of 4-7% for gastroschisis [19,20] and 8-27% for omphalocele [21,22]. Comparison of mortality shows significative differences between low- and hight-income countries, which is mostly the result of the level of development of their health care system (table IV). Conversely, development of surgical techniques, advances in surgical and neonatal care have improved survival of neonates with omphalocele, challenges with respiratory failure, prolonged intensive care stay, poor feeding and growth, and neurodevelopmental delay remain significant risks [21,25]. The optimal postoperative feeding regimen in infants with gastroschisis is currently debate. In this study we perform partial parenteral nutrition and fractional feeding on breast milk exclusively. Wright et al report that only 19% of low income countries surgeons had access to parenteral nutrition compared with 100% of those of high income countries [19]. For Storm et al, mother's own milk dose was significantly associated with a decrease in time to discharge from the initiation of enteral feedings and lenght of hospital stay [27]. Early enteral feeding, but with

slow progression, has been proven to be beneficial in patients with gastroschisis. It's shortens the duration of total parenteral nutrition, hospital stay and may help decrease the rate of sepsis [28]. This will be especially important in our setting where parenteral nutrition is not readily available.

In this series, the main cause of death was sepsis such as in the study of Oyinloye et al at Nigeria [16]. That could be explained by multiple manipulations, delay for surgery and the fact that none cases of eviscerate bowel or omphalocele bag were protected sterily before admission. The neonate is fragile with a weak immune status and body systems not quite adapted to the extra uterine environment, and therefore very susceptible to infections which can occur both locally and generally. Thus, our babies were transported in poor conditions without care defined by the "STABLE program″ which emphasises : Sugar control, Temperature control, Airway maintenance, Blood pressure, Laboratory work and Emotional support of the family [29]. Anaemia was the second cause of death. This anaemia needed blood transfusion, but it was not corrected due to the frequent dearth of blood products in our practice. Another cause of death was respiratory distress. Early closure of the abdominal wall may lead to a sudden increase in intra-abdominal pressure and respiratory insufficiency due to diminished lung excursion. Facilities of monitoring and mecanic ventilators were unavailable in our unit. The role of lung hypoplasia and associated malformation in the death was not studied. In general, CAWD lead to psychological, affect the quality of life of these neonates and their families.

Table IV: Comparative table of mortality in our series with those of others series

	Number of cases (N)	Mortality n(%)	p
Omphalocele			
Côte d'Ivoire (our series)	129	40 (31)	
Senegal [23]	95	43 (45,3)	0.142
Japon [22]	399	80 (20)	0.045
USA [24]	274	52 (19)	0.036
Netherland [25]	69	17 (24,6)	0.480
Korea [2]	60	9 (15)	0.066
Gastroschisis			
Côte d'Ivoire (our series)	30	26 (86,6)	
Ouganda [7]	42	41(98)	0.731
Nigeria [8]	39	34 (87,2)	0.987
Bangladesh [14]	75	61(81,3)	0.842
Japon [26]	247	20 (8,1)	0.000
USA [20]	566	30(5,3)	0.000

Conclusion

Congenital anterior abdominal wall defects are characterized by a marginal prenatal diagnosis. They still pose a challenge in our practice with a high mortality rate. Improvement of the technical platform, effective health insurance, popularization and promotion of neonatal primary care in our hospitals, equal distribution of pediatric surgeon, will improve the prognosis of this malformation.

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Conflict interest : None

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