Anterior Abdominal Wall Defects, Diaphragmatic Hernia, and Other Major Congenital Malformations of the Musculoskeletal System in Barbados, 1993–2012

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Abstract

Keywords

- Caribbean
- congenital malformations
- musculoskeletal
- prevalence
- mortality

This study describes the prevalence and patterns of major congenital malformations of the musculoskeletal system and the resulting morbidity and mortality. It is a retrospective population-based study over the period 1993 to 2012. The overall prevalence of major congenital malformations of the musculoskeletal system was 9.02/10,000 live births. The prevalences of omphalocele, gastroschisis, and diaphragmatic hernia were 2.53, 2.22, and 1.42 per 10,000 live births, respectively. The case fatality ratio for the omphalocele, gastroschisis, and diaphragmatic hernia was 12.5, 28.5, and 67%, respectively. In conclusion, the prevalence rate of the major congenital malformations of the musculoskeletal system was higher than those reported in retrospective studies from other countries and remained static during the study period. These defects were associated with a high mortality rate and contributed significantly to the overall neonatal mortality in this country.

Introduction

The musculoskeletal system is reported to be one of the more commonly affected systems with major congenital malformations (International Classification of Diseases [ICD] class Q65–Q79) in many parts of the world including Barbados.^{1–5} Osteochondrodysplasias (Q77 and Q78) are reported to be the most common major malformation affecting the musculoskeletal system and most of them are incompatible with life.^{6,7} Among other major congenital malformations of the musculoskeletal system (MCMMS) not elsewhere classified (Q79), that is, abdominal wall defects, and diaphragmatic hernia, are more common and life-threatening in the neonatal period. Omphalocele and gastroschisis are the two major malformations of the abdominal wall with the reported combined prevalence varying between 3 and 5 per 10,000 live births.^{8–12} Omphalocele is

reported to be more prevalent than gastroschisis.^{8–12} Although the birth prevalence of omphalocele has remained generally stable over past 20 years, there are reports suggesting a 10-fold increase in the rate of gastroschisis over the past decade.^{11–15} Both omphalocele and gastroschisis carry significant neonatal mortality with reported rate as high as 22 and 9%, respectively.^{16,17} Diaphragmatic hernia, one of the other more common major musculoskeletal malformations, has a reported prevalence of 3 to 4 per 10,000 live births.^{18,19} It also carries a high mortality with the reported rate as high as 43%.²⁰ The prevalence and patterns of major malformations of the musculoskeletal system are reported to have geographic, ethnic, and temporal variations.^{8–15,19,20} There are no data published till date from the English-speaking Caribbean on the prevalence and patterns of MCMMS.

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Barbados, one of the English-speaking Caribbean countries has a total population of 283,000 (2012) including 58,500 (21%) children under the age of 16 years.²¹ Its gross national income per capita (PPP international \$) is US\$15090 (2012) and its total expenditure on health as a percentage of GDP stands at 6.3%.²¹ The infant mortality rate (2005) is 10.93 per 1,000 live births.²² The antenatal follow-up for pregnant women is more than 90%.²³ There is a central birthing facility at the Queen Elizabeth Hospital (QEH), the sole tertiary care hospital for the entire population, and this provides a unique opportunity to study the congenital malformations in this population. In this study, we describe the prevalence and patterns of major malformations of the musculoskeletal system in this country and its time trend. We also describe the morbidity and mortality from these malformations.

Material and Methods

This is a retrospective study of babies born with MCMMS from 1993 to 2012. Cases were identified from the admissions register maintained in the neonatal intensive care unit (NICU) at the QEH. Still births with congenital malformations were excluded from this study. Data on the babies with MCMMS were obtained from the birth register in the labor ward and the admissions register at the NICU. This included date of delivery, maternal age, maternal parity, gestational age at delivery, birth weight, and gender. Additional population data such as total number of live births, number of neonatal admissions, total number of babies born with major congenital malformations, death among babies with any major malformations and number of deaths in babies with MCMMS, and overall total number of neonatal deaths during the study period were also collected from these registers. The hospital case records for all the babies with MCMMS were also reviewed to collect additional data on the neonates and their mother. Maternal data included history of trauma, infection, chronic diseases, and the use of alcohol or drugs in pregnancy. Baby's data that included other associated malformations and outcome in terms of hospital discharge or death were also collected. Liveborn infants were examined after birth by a pediatrician, and congenital anomalies, if any, were described in detail.

We classified the study group according to the ICD-10 classification of diseases by the World Health Organization. Under this classification, diaphragmatic hernia (Q79.0), gastroschisis excluding umbilical hernia (Q79.3), and omphalocele (Q79.2) fall under the category of congenital malformations of the musculoskeletal system not elsewhere classified (Q79). Omphalocele was defined as evisceration with a covering membrane and the umbilical cord present within the sac. Gastroschisis was defined as evisceration without a covering membrane and the umbilical cord in the abdominal wall apart from the defect. Prematurity was defined as delivery less than 37 complete weeks of gestation. Small for gestational age (SGA) was defined as the birth weight below the 10th percentile for the given gestational age.

Ethical approval was obtained from the ethics committee at the QEH and Institutional Review Board for ethics in study involving human subjects of the University of the West Indies and the Ministry of Health, Barbados. To protect the personal

information of the patients, only the investigators had access to the database, and the data were stored in a specially created Microsoft Excel spreadsheet with password. The same software program was used for data tabulation and generation of graphs. The results were analyzed by simple statistical techniques, and tests of significance including chi-squared test were applied.

Results

During the 20-year study period, there were a total of 57 cases of MCMMS (Q65–Q79) from among the 63,176 live births. Omphalocele was the single most common major malformation with 16 (28.0%) cases. This was followed by gastroschisis 14 (24.6%) cases, diaphragmatic hernias in 9 (15.8%) cases, and others in 18 (31.58%) cases. All the nine cases of diaphragmatic hernias were of the posterolateral types. Trends in the prevalence rate of MCMMS along with the number of live births during the study period are shown in ►Fig. 1. The overall prevalence rate of MCMMS (Q65–Q79) was 9.02 (95% confidence interval [CI] = 7.95–9.59) per 10,000 live births. The overall prevalence rate for omphalocele, the most common form of MCMMS, was at 2.53 (95% CI = 1.56–3.81) per 10,000 live births over the study period, whereas that of gastroschisis was 2.22 (95% CI = 1.31–3.47) per 10,000 live births. The difference in the overall prevalence rate of omphalocele and gastroschisis was not statistically significant ($p = 0.061$).

Among other major musculoskeletal malformations, there were seven cases of congenital malformations of the spine and bony thorax (Q76), five cases of osteochondrodysplasia with defects of growth of tubular bones and spine (Q77), three cases of congenital malformations of the musculoskeletal system not elsewhere classified (Q79), including two cases of prune belly syndrome (Q79.4) and one case of other congenital malformation of the anterior abdominal wall (Q79.5), two cases each of the other osteochondrodysplasias, unspecified (Q78.9), and one case each of the reduction defects of unspecified limb (Q73.1), other reduction defects of right upper limb (Q71.8), other congenital musculoskeletal deformities (Q68.2), and other congenital deformities of skull face and jaw (Q67.4). The prevalence rate for severe forms of osteochondrodysplasias in this study was 2 per 10,000 live births.

Selected demographic characteristics of the neonates born with MCMMS are shown in ►Table 1. Overall, majority of the neonates with MCMMS were born to mothers in the age group of 21 to 35 years ($n = 39$, 68.4%), at the gestational age group of 37 to 40 weeks ($n = 31$, 54.4%), and with a birth weight between 2,500 and 4,000 g ($n = 35$; 61.4%). Analysis of the actual prevalence rate revealed that MCMMS were most prevalent among mothers in the age group of 21 to 35 years (9.62/10,000 live births), in the gestational age group of <37 completed weeks (14.14/10,000 live births), and in the birth weight category of <2,500 g (15.82/10,000 live births). However, the difference in prevalence rate was statistically significant only for the gestational age groups ($p = 0.03$) and the birth weight categories ($p = 0.002$). The risk of having a MCMMS was significantly higher ($p = 0.017$) in the babies born at term gestation (relative risk [RR] = 2.34). Omphalocele was most prevalent among mothers in the age group of

*Total number of live births in Barbados during the study period were as follows:

1993-1997	15692
1998-2002	15966
2003-2007	16284
2008-2012	15234

Fig. 1 Trends in the prevalence rate* of the major congenital malformations of the musculoskeletal system in Barbados, 1993 to 2012.

Table 1 Prevalence (per 10,000 live births) of various MCMMS among various maternal age group, gestation age group, birth weight group, and gender in Barbados, 1993 to 2012

	All	Omphalocele	Gastroschisis	Diaphragmatic hernia
Age group				
< 21 (n = 10,743)	10 (9.31)	1(0.93)	7 (6.52)	1 (0.93)
21-35 (n = 40,545)	39 (9.62)	12 (2.96)	6 (1.48)	7 (1.73)
36-50 (n = 11,888)	6 (5.05)	2 (1.68)		1 (0.84)
No data	2 (0.31)	1 (0.15)	1 (0.15)	4 (0.62)
Gestational age				
< 37 (n = 8,487)	12 (14.14)	2 (2.36)	5 (5.89)	3 (3.53)
37-40 (n = 49,946)	31 (6.21)	7 (1.40)	2 (0.40)	4 (0.80)
> 40 (n = 4,743)	2 (4.22)	3(6.33)	2 (4.22)	2 (4.22)
No data	12 (1.90)	4 (0.63)	5 (0.79)	21 (3.32)
Birth weight				
<2,500 g (n = 12,645)	20 (15.82)	4 (3.16)	9 (7.12)	1 (0.79)
2,500-4,000 g (n = 40,409)	35 (8.66)	11 (2.72)	5 (1.24)	7 (1.73)
>4,000 g (n = 10,122)	2 (1.98)	1 (0.99)	0 (0.0)	1 (0.99)
Gender				
Female (n = 32,316)	29 (8.97)	6 (1.86)	9 (2.78)	5 (1.55)
Male (n = 30,860)	27 (8.75)	9 (2.92)	5 (1.62)	4 (1.30)
No data	1 (0.15)	1 (0.15)	0	2 (0.30)

Abbreviation: MCMMS, major congenital malformations of the musculoskeletal system.

21 to 35 years (2.96/10,000 live births), in the gestational age group of >40 weeks (6.33/10,000 live births), and in the birth weight category of <2,500 g (3.16/10,000 live births). There was a slight male preponderance (2.92/10,000 live births)

among the cases of Omphalocele. However, none of these differences in the prevalence rates among various categories was statistically significant. Gastroschisis was significantly more prevalent among mothers in the age group of <21 years

Table 2 MCMMS during the 1993 to 2012 period in Barbados

Morbidity and mortality indicators	All other causes	MCMMS
Admission to NICU	11,785 (99.5%)	57 (0.5%)
Neonatal deaths	600 (97.2%)	17 (2.8%)

Abbreviations: MCMMS, major congenital malformations of the musculoskeletal system; NICU, neonatal intensive care unit.

(6.52/10,000 live births; $RR = 5.7$; $p = 0.0012$) and in the gestational age group of <37 weeks (5.89/10,000 live births; $RR = 8.1$; $p = 0.0001$), with a slight female preponderance (2.78/10,000 live births). Diaphragmatic hernia was most prevalent among mothers in the age group of 21 to 35 years (1.73/10,000 live births), in the gestational age group of >40 weeks (4.22/10,000 live births), and in the birth weight category of 2,500 to 4,000 g (1.73/10,000 live births). Only the difference in the prevalence among various maternal age groups was statistically significant ($p = 0.035$).

Overall, five mothers had diabetes mellitus. Three had diabetes mellitus diagnosed prior to their pregnancy and were on insulin with fairly good control during their pregnancy. Two other women were diagnosed with gestational diabetes and maintained control on insulin. Illicit drug abuse were self-reported by four mothers, of whom three smoked marijuana and one used inhaled cocaine, all of whom had a baby with gastroschisis. Two mothers had epilepsy, both controlled on carbamazepine. One mother had systemic lupus erythematosus in remission, and another mother had hypothyroidism controlled on thyroxine.

Time trends in the prevalence of the different musculoskeletal malformations over the 20 years period are shown in ► **Fig. 1**. The mean annual incidence rate of MCMMS during the study period was 9.05 (SD = 1.10) per 10,000 live births. The mean annual incidence of MCMMS varied between 7.37 per 10,000 live births (2003–2007) and 10.20 per 10,000 live births (1993–1997). No significant trends were observed. The prevalence rate of gastroschisis decreased from 4.46 in 1993 to 1997 to 1.84 in 2003 to 2007 per 10,000 live births, and no cases were seen during 2008 to 2012. However, this decrease in the prevalence rate over time was statistically not significant ($p = 0.06$). No significant time trend was observed in the prevalence of omphalocele. The prevalence rate for diaphragmatic hernia increased during the study period but no significant trend was noted.

There were a total of 11,842 neonatal admissions during the 20 years study period. Overall, neonates with MCMMS were responsible for 0.48% (95% CI = 0.37–0.63%) of the total neonatal admissions (► **Table 2**). The average duration of the NICU stay was 23 days compared with the average stay of 16 days for all the NICU admissions. Among babies born with omphalocele, in 43.7% (7/16) of cases, it was an isolated malformation, whereas in the rest of the cases, there were one or more other associated malformations. Two cases had club foot deformity, two had acyanotic heart disease with ventricular septal defects, and one each had hypospadias, small bowel obstruction, and right polydactyly with preaxial thumb. Among the babies born with gastroschisis, in 78.5% (11/14) of cases, it was an isolated malformation, and in the rest of the cases, there were one or more other associated

Table 3 Case fatality ration for the MCMMS during the 1993 to 2012 period in Barbados

Malformations of the musculoskeletal system	Deaths (case fatality ratio)
Omphalocele ($n = 16$)	2 (12.5%)
Gastroschisis ($n = 14$)	4 (28.6%)
Diaphragmatic hernias ($n = 9$)	6 (67%)
All MCMMS ($n = 57$)	17 (29.8%)

Abbreviation: MCMMS, major congenital malformations of the musculoskeletal system.

malformations. In two cases, there was an acyanotic congenital heart disease (one ventricular septal defect and one pulmonary stenosis). Among the babies born with diaphragmatic hernias, in 44.4% (4/9) of cases, there were one or more other associated malformations. One each had cleft lip, spinabifida occulta, ventricular septal defect, and club foot. None of the babies had any chromosomal study done. Of the 617 neonatal deaths during the study period, 17 (2.75%; 95% CI = 1.67–4.47%) deaths were attributed to MCMMS. The overall case fatality ratio (CFR) for neonates with MCMMS was 29.80%. Highest CFR (67%) was seen among neonates with diaphragmatic hernia (► **Table 3**).

Discussion

Among congenital malformations of the musculoskeletal system, not elsewhere classified, the abdominal wall defects (including omphalocele and gastroschisis) and the diaphragmatic hernias constitute the three most common life-threatening major congenital malformation seen at birth.^{8–12,18,19,24} Omphalocele or exomphalos refers to a protrusion of abdominal content into the umbilicus. In its mild form, a small loop of intestines protrudes into the base of the umbilicus, which is a hernia into the umbilical cord. Its more severe form constitutes an omphalocele in which a midline anterior abdominal wall defect allows protrusion of small intestine and other viscera directly into the umbilical cord, thereby distending its base into a cystic mass containing viscera. Gastroschisis is a defect lateral to the midline in the full anterior abdominal wall through which the abdominal contents protrude directly into the amniotic cavity.²⁵ Diaphragmatic hernia is a defect in the diaphragm, more often on the left hemidiaphragm than the right hemidiaphragm, with herniation of the abdominal contents into the thorax during the intrauterine life.²⁶

In this first published study from the English Caribbean, we report on the prevalence and pattern of MCMMS, with a particular focus on omphalocele, gastroschisis, and diaphragmatic hernias, from a passive surveillance in a predominantly black population of Afro-Caribbean descent. The overall prevalence of MCMMS during the entire study period was 9.02 per 10,000 live births. Several retrospective, mostly hospital-based, studies from countries from around the world have reported a lower prevalence rates of MCMMS compared with ours.^{1–3,9,11,27–30}

However, a much higher prevalence rate (62/10,000 births) of MCMMS compared with ours have been reported from Europe which has a well-established population-based prospective passive surveillance system with an established birth defect registry (EUROCAT).³¹ Higher prevalence seen in the EUROCAT report may be reflective of the better data collection and capture.

In this population, MCMMS constituted 15.5% of all major congenital malformations seen at birth, which is once again similar to those reported from several other countries around the world.^{1–3,27–30} Omphalocele (28.07%), gastroschisis (24.56%), and diaphragmatic hernia (15.79%) together constituted over four-fifths of all MCMMS in this setting. In this study, omphalocele (2.56/10,000 live births) was more prevalent than the gastroschisis (2.22/10,000 live births), although the difference in the prevalence was statistically not significant. Omphalocele has been reported to be more common than gastroschisis in several other studies from Asia and Europe.^{8–10,27} Other patterns of MCMMS constituted less than a fifth of the total MCMMS and were mostly (a total of 14 cases) osteochondrodysplasias including skeletal dysplasias and defects of growth of tubular bones and spine. The overall prevalence rate for the severe forms of osteochondrodysplasia was 2/10,000 live births, which is comparable with other reported studies.^{6,7} Four cases were labeled as thanatophoric dysplasia, two as asphyxiating thoracic dystrophy, and two as osteogenesis imperfecta type 2. This group of MCMMS had a high mortality rate, with 10 out of 14 cases dying in the neonatal period.

Overall, MCMMS were significantly more prevalent among the premature (<37 weeks) and low birth weight babies. Omphaloceles were more prevalent among mothers in the age group of 21 to 35 years, postterm, and low birth weight babies; however, the differences were statistically not significant. Gastroschisis was significantly more prevalent among younger mothers (<21 years), preterm delivery, and low birth weight babies. Similar findings have been reported in several published studies.^{9,11,12,14} Babies with omphalocele and gastroschisis were both significantly more likely to be SGA. Once again, similar findings have been reported in other studies.^{9,11,12,14} Diaphragmatic hernia was more prevalent among mothers in the age group of 21 to 35 years and among babies born at gestational age 37 to 40 and with birth weight 2,500–4,000 g. These findings are similar to those reported in literature.^{18–20} During the 20-year study period, incidence rate of omphalocele was stable, whereas the incidence rate of gastroschisis declined, although this decline was statistically not significant. Several studies from Asia, Europe, and North America have shown an increase in the incidence rate of gastroschisis in recent years.^{11–14} The decline in the prevalence of the gastroschisis seen in this study may have been due to more frequent antenatal ultrasound examination in the recent years with antenatal detection of these defects and termination of pregnancy, which have not been captured in this study.

Nearly half of omphalocele and four-fifths of gastroschisis cases were isolated malformations. Majority of the associated malformations in either case were musculoskeletal or cardiac. Similar findings have been reported in other studies.^{9,10,14,32} Babies with MCMMS during the study period had a signifi-

cantly higher mortality compared with the overall neonatal mortality rate, with nearly one-third of these babies dying during the neonatal period. One-eighth of all cases of omphalocele died during the neonatal period, which is similar to those reported in other studies.^{11,15} Just over a fourth of gastroschisis died during the neonatal period, and this was higher than those reported in other studies.^{11,15}

The mortality from MCMMS (17 deaths) accounted for 2.7% of the overall neonatal mortality (617 neonatal deaths) during the study period. However, the contribution of these malformations to the overall neonatal mortality has to be seen against the changing pattern of the neonatal morbidity and mortality in this country. During the 1990s and the early 2000s, birth-related trauma, neonatal sepsis, and prematurity contributed significantly to the neonatal morbidity and mortality.³³ In recent years, there has been decreasing mortality from neonatal sepsis and birth trauma (data from the annual perinatal morbidity and mortality review, 2013, 2014, and 2015). Therefore, the mortality from malformation including those of the musculoskeletal system in the coming years is expected to increase, all other things being the same. Our mortality rates for both omphalocele and gastroschisis were higher than those reported in recent studies from other developed countries.^{25,34} Mortality was particularly higher among babies with diaphragmatic hernias, and a little more than two-thirds of these babies died during the neonatal period. This is much higher than those reported in recent series with advanced perioperative supportive care.^{35,36} Lack of adequate intensive supportive care in the perioperative period was responsible for this high mortality, which is similar to those reported in series in the nineties.^{37,38}

In conclusion, findings from this study for the first time provide data on MCMMS from the English-Speaking Caribbean. The prevalence rate of the various MCMMS seen in this population was higher than those reported from other retrospective, mostly hospital-based studies from around the world. However, a much higher prevalence rate is reported in studies where the data was collected prospective by way of well-established birth defect registry and makes a strong case for establishment of a birth defect registry in this region. Majority of the cases were detected at the time of delivery. Compared with recent reports from the developed countries, we had a very high mortality rate for all three MCMMS. Routine antenatal screening for these defects may provide the option of terminating those cases with associated other potentially lethal malformations, and better preparation for management of other cases. This may help in reducing the mortality rate for these birth defects.

Conflict of Interest

None.

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