

Original Article

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Submitted: 04-03-2023 **Accepted:** 04-04-2023

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DOI: https://doi.org/10.47338/jns.v12.1199

Epidemiology of congenital diaphragmatic hernia, esophageal atresia, and gastroschisis in South Brazil

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KEYWORDS

Gastroschisis

Congenital abnormalities, Congenital Diaphragmatic Hernias, Esophageal Atresia, Epidemiology,

ABSTRACT

Background: Birth defects remain a major contributor to infant mortality and lifelong disabilities. The epidemiology of congenital abnormalities varies around the world and little information is available from Latin America.

Methods: This is an epidemiological, descriptive, cross-sectional study with data from the Department of Informatics of the Unified Health System (DATASUS) on Congenital diaphragmatic hernia (CDH), esophageal atresia (EA) and gastroschisis (GS) in South Brazil from 2009 to 2019.

Results: The incidence of CDH is 0.93 cases, while EA is 0.47 and GS is 2.87, all per 10,000 live births. There is an association between all the malformations and premature birth, cesarean delivery, low birth weight, and low Apgar scores. Both EA and GS are associated with maternal age, EA with older, and GS with younger mothers. While EA is associated with multiple pregnancies, GS is associated with fewer years of maternal formal education, single parenting, and a lower number of prenatal consultations. CDH is associated with the male sex and black ethnicity.

Conclusion: This large population-based study estimates the prevalence and demographic factors associated with CDH, EA, and GS, and extends the limited descriptive epidemiologic information available.

INTRODUCTION

Birth defects are congenital structural or genetic conditions that cause significant health and developmental complications. They remain a major contributor to infant mortality and lifelong disabilities. [1] An estimated 240,000 newborns die worldwide within 28 days of birth every year due to birth defects, and they cause a further 170,000 deaths of children between the ages of 1 month and 5 years. [2]

Although nine of ten children born with a serious birth defect are in low- and middle-income countries, data from these regions are sparse because the databases commonly do not reach all deaths or do not include some essential information. [2, 3] The true human and financial cost of congenital anomalies remains grossly underestimated. [4]

Brazil lacks a national population-based surveillance program to track major birth defects, but Datasus offers all this data, just in a raw form. A better understanding of the epidemiology of these diseases might assist clinicians and policymakers to make the provision of adequate care a priority, improving the well-being of billions of children. [5]

This paper aims to describe the epidemiology of congenital diaphragmatic hernia (CDH), esophageal atresia (EA), and gastroschisis (GS) in South Brazil (Paraná, Santa Catarina, and Rio Grande do Sul) from 2009 to 2019.

METHODS

This is an epidemiological, descriptive, cross-sectional study with data from the Department of Informatics of the Unified Health System (DATASUS) on CDH, EA, and GS from 2009 to 2019. Data were accessed on the Live Birth Information System (SINASC) website. [6]

We analyzed maternal age and years of formal education, marital status, number of prenatal consultations, type of pregnancy (singleton or multiple), gestational age, mode of delivery, birth weight (BW), sex, ethnicity, and Apgar scores at 1 (Apgar 1) and 5 (Apgar 5) minutes.

Descriptive statistics were used to analyze the quantitative portion of the study, with results presented as percentages, means, and medians. Fisher's Exact Test was used to evaluate categorical variables, and Bonferroni correction was applied for multiple comparisons. Analyses were conducted with R, version 3.5.0 (R Core Team, Vienna, Austria), and the statistical difference was considered when p value ≤0.05.

RESULTS

Between January 2009 and December 2019, there were 4,255,556 live births (LB) in South Brazil: 1,705,370 in Paraná, 1,015,983 in Santa Catarina and 1,534,203 in Rio Grande do Sul. The total number of CDH, EA, and gastroschisis cases in this period can be seen in Figure 1/Table 1 and the analyzed parameters regarding each malformation are found in Table 2.

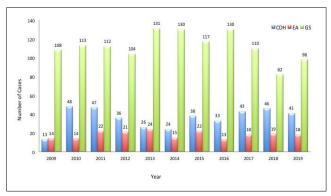


Figure 1. The number of Congenital Diaphragmatic Hernia (CDH), Esophageal Atresia (EA), and Gastroschisis (GS) cases in South Brazil per year, 2009-2019.

Table 1. The total number of Congenital Diaphragmatic Hernia (CDH), Esophageal Atresia (EA), and Gastroschisis (GS) cases in South Brazil, 2009-2019

	CDH	EA	GS	Total Cases	Live Births	
PR	150	81	466	697	1,705,370	
sc	96	49	305	450	1,015,983	
RS	149	70	454	673	1,534,203	
Total	395	200	1225	1820	4,255,556	
Prevalence per 10,000 births	0.92	0.46	2.87	4.27	-	

DISCUSSION

Congenital diaphragmatic hernia (CDH): CDH refers to a defect of the diaphragm formation that often presents in the neonatal period with moderate to severe respiratory distress. [7]

CDH has a reported incidence that ranges between 0.7 and 2.61 in 10,000 live births, depending on the

geographical area and the period examined. [8–10] From 2009 to 2019, 395 CDH cases were reported in South Brazil, with an incidence of 0.93 cases per 10,000 live births. The highest incidence was in Rio Grande do Sul (0.97). Santa Catarina was in second (0.94), and the lowest incidence was in Paraná (0.87)

The mean maternal age in CDH cases was 27.8 years and it was 27.3 among non-CDH mothers. There was no association between maternal age and CDH (p=0.38). Our findings were similar to others that did not find any association between maternal age with CDH. [3, 11] On the other hand, a systematic review from Paoletti et al. [10] found that maternal age >35 years was significantly associated with the disease, but the age evaluation analyzed only five papers, none of them from South America. Yang et al. [12] found relative risks were elevated by at least 50% in maternal age >35 years, but only for non-isolated CDH cases.

Most CDH mothers had eight or more years of formal education (n=318/394, 80.7%), were married or in a stable relationship (n=216/389, 55.5%), had seven or more prenatal consultations (n=301/392, 76.8%) and had single pregnancy (n=385/395, 97.5%). No association was found between all these variables and the presence of CDH (Table 2).

There was an association between CDH and birth <37 weeks (p<0.01), and cesarean delivery (p<0.01).

The mean birth weight (BW) for CDH babies was 2794.6 g, while for non-CDH babies was 3189.2 g, there was an association between CDH and lower BW (p<0.01).

The median Apgar 1 and Apgar 5 in CDH cases was 5 and 9, respectively, and in non-CDH babies was 7 and 10. This was also statistically significant (p<0.01).

CDH babies are born at a significantly earlier gestational age (GA), with lower BW and Apgar scores. [3, 11]

Although neither mode nor time of delivery seems to affect the outcome for patients with prenatally diagnosed CDH, [13] children born with this malformation are significantly more often delivered by cesarean section. [11] In this study, we did find an association between surgical delivery and CDH.

There was also an association between CDH and the male sex (p=0.02). Although some studies did not find differences in gender distribution, [11] our finding that males are more likely to have CDH is in general agreement with previous studies. [10, 12, 14] According to Woodbury et al., [14] the rate ratio for males is 1.5.

While some papers did not find an association between maternal ethnicity and CDH, [11] others showed that babies of black descent were less likely to develop the malformation. [10, 12, 14] On the contrary, a previous study from São Paulo state (Brazil) reported a higher prevalence of CDH in babies from black mothers than from other ethnic categories. [3] We found an association between black ethnicity

and CDH (when comparing black and non-caucasians; p=0.03) with an incidence of 1.49 cases per 10,000 live births within this subgroup, which could be a peculiarity of our country/region.

Table 2. Characteristics of Live Births (LB) with Congenital Diaphragmatic Hernia (CDH), Esophageal Atresia (EA), and Gastroschisis (GS) in South Brazil, 2009-2019

Characteristics	LB	CD	CDH		EA		GS	
		n (%)	р	n (%)	р	n (%)	р	
Maternal age								
<= 19	654,849	47 (11.90)	0.38	37 (18.5)	<0.01 ^b	578 (47.18)	<0.01°	
20-34	2983777	278 (70.38)		119 (59.5)		628 (51.27)		
>= 35	616607	70 (17.72)		44 (22)		19 (1.55)		
Ignored	323	0		0 (0)		0 (0)		
Maternal schooling								
0-7 years	921072	76 (19.24)	0.53	45 (22.5)	0.47	337 (27.51)	<0.01	
>= 8 years	3316049	318 (80.51)		155 (77.5)		881 (71.92)		
Ignored	18435	1 (0)		0 (0)		7 (0.57)		
Marital status								
Single	1898243	173 (43.80)	0.67	86 (43)	0.50	750 (61.23)	<0.01 ^d	
Married/stable union	2266905	216 (54.68)		108 (54)		457 (37.31)		
Widowed/divorced	67114	6 (1.52)		5 (2.5)		9 (0.73)		
Ignored	23294	0 (0)		1 (0.5)		9 (0.73)		
Consultations								
0-6	973286	91 (23.04)	0.94	59 (29.5)	0.07	463 (37.80)	<0.01	
7 ou +	3266350	301 (76.20)		139 (69.5)		756 (61.71)		
Ignored	15920	3 (0.76)		2 (1)		6 (0.49)		
Pregnancy								
Singleton	4156625	385 (97.47)	0.57	188 (94)	0.04	1206 (98.45)	0.46	
Multiple	96407	10 (2.53)		12 (6)		18 (1.47)		
Ignored	2524	0 (0)		0 (0)		1 (0.08)		
Gestational age								
< 37 weeks	424000	117 (29.62)	<0.01	87 (43.5)	<0.01	613 (50.04)	<0.01	
>= 37 weeks	3648549	264 (66.84)		98 (49)		537 (43.84)		
Ignored	183007	14 (3.54)		15 (7.5)		75 (6.12)		
Delivery								
Vaginal	1676311	62 (15.70)	<0.01	53 (26.5)	<0,01	208 (16.98)	<0.01	
Cesarian	2577202	332 (84.05)		147 (73.5)		1016 (82.94)		
Ignored	2043	1 (0.25)		0 (0)		1 (0.08)		
Birth weight								
> 2500 g	372942	110 (27.85)	<0.01	92 (46)	0.01	787 (64.24)	<0.01	
>= 2500 g	3878738	285 (72.15)		107 (53.5)		438 (35.76)		
Ignored	3876	0 (0)		1 (0.5)		0 (0)		

Apgar 1							
0-6	277128	270 (68.35)	<0.01	61 (30.5)	<0.01	399 (32.57)	<0.01
7-10	3977253	125 (31.65)		139 (69.5)		826 (67.43)	
Ignored	1175	0 (0)		0		0 (0)	
Apgar 5							
0-3	66530	151 (38.23)	<0.01	20 (10)	<0.01	99 (8,08)	<0,01
7-10	4187943	244 (61.77)		180 (90)		1126 (91,92)	
Ignored	1083	0 (0)		0 (0)		0 (0)	
Sex							
Male	1978387	222 (56.20)	0.02	106 (53)	0.12	549 (44.82)	0.10
Female	1880333	149 (37.72)		79 (39.5)		546 (44.57)	
Ignored	396836	24 (6.08)		15 (7.5)		130 (10.61)	
Race							
White	3532286	333 (84.30)	0.03 ^a	166 (83)	0.76	1016 (82.94)	0.96
Black	160344	24 (6.08)		8 (4)		50 (4.08)	
Others	519808	36 (9.11)		24 (12)		149 (12.16)	
Ignored	43118	2 (0.51)		2 (1)		10 (0.82)	

Bonferroni correction for multiple comparisons: a Black vs white p=0.06; black vs others p=0.03; white vs others p=0.99. b Mothers \leq 19 years vs 20-35 years p=0.87; \leq 19 years vs \geq 35 years p=0.15; 20-35 years vs \geq 35 years p<0.01. c Mothers \leq 19 years vs 20-35 years p<0.01; \leq 19 years vs \geq 35 years p<0.01; \leq 19 years vs \geq 35 years p<0.01; d Single mothers vs married/stable union p<0.01; single mothers vs widowed/divorced p=0.04; married/stable union vs widowed/divorced p=0.99.

Esophageal atresia (EA): EA is the most frequent anomaly of the esophagus and is characterized by a congenital esophageal disruption with or without tracheoesophageal fistula. [15, 16] Diagnosis can be formed during the prenatal scans or, in most cases, at birth, and surgical repair is required in the first few days of life. [16] Its incidence is reported to vary from 0.7 to 4.53 per 10,000 births. [15–18]

From 2009 to 2019, 200 EA cases were reported in South Brazil. The total incidence was 0.47 cases per 10,000 live births, and it was very similar among all three states (0.48 in Santa Catarina, 0.47 in Paraná and 0.46 in Rio Grande do Sul, all per 10,000 live births).

This lower incidence found in our study (a little under 0.5 cases per 10,000 LB) could be the true incidence in South Brazil, but could also be due to some study limitations: (1) only live births were included in this study; (2) some publications considered esophageal stenosis as EA; [16] (3) the association between EA and other malformations is very common and these cases might have been placed under a different diagnosis in DATASUS; [16, 18, 19] (4) Brazilian protocols recommend that the orogastric catheter is not used in the delivery room, which could delay the diagnosis, and, since our data was based on delivery room information, some cases might have been missed. [20]

The mean maternal age in EA cases was 28.02 years and it was 27.27 among non-EA mothers, and there was an association between EA and mothers ≥ 35 years when compared to mothers 20-35 years (p<0.01). Our findings are supported by other studies that described an elevated relative risk for mothers >35 years, although this is not a consensus. [16, 17]

Most EA mothers had eight or more years of formal education (n=155/200; 77.5%), were married or in a stable relationship (n=108/199; 54.3%), and had seven or more prenatal consultations (n=139/198; 70.2%). No association was found between all these variables and the presence of EA.

There was an association between EA and multiple pregnancies (p=0.04), birth <37 weeks (p<0.01), and cesarean delivery (p<0.01). The association between twin pregnancy and EA has been previously reported, and mortality is greater in this subgroup. [16, 18, 21, 22] Although there is no proven benefit regarding the mode of delivery, [23] cesarean was more commonly performed in South Brazil (n=149; 73,76%), and we found an association between EA and surgical delivery (p<0,01, Table 2). This finding is supported by previous studies. [24]

The mean BW for EA babies was 2441.96 g, while for non-EA babies was 3189.24 g, and there was an association between EA and lower BW (p<0.01). Previous reports also described an association

between lower GA and EA. [18, 19] The association between low BW and EA is well established, and this association was also found in our study. There is speculation that the high mechanical obstruction seen in EA may lead to intrauterine growth retardation from decreased absorption of amniotic fluid since its proteins are believed to be absorbed by fetal intestines and used in fetal protein synthesis. [17–19]

The median Apgar 1 and Apgar 5 in EA cases was 8 and 9, respectively, and in non-EA babies was 9 and 10. This difference was statistically significant (p<0.01).

Although the majority of neonates were male (n=108, 53,46%) and previous papers described significant differences in EA incidence between races, we found no statistical significance regarding sex (p=0.12) or race (p=0.76). [16–18]

Gastroschisis (GS): GS is a congenital anomaly of the anterior abdominal wall associated with bowel evisceration that requires surgical correction and is generally associated with prolonged hospitalization, high costs, and high neonatal morbidity. [25, 26]

From 2009 to 2019, 1,225 GS cases were reported in South Brazil, with an incidence of 2.87 cases per 10,000 live births. The highest incidence was in Santa Catarina (3.01); Rio Grande do Sul was in second (2.96), and the lowest incidence was in Paraná (2.73).

Worldwide, GS incidence varies from 1.1 cases to 4.49/10,000 live births. [9, 27, 28] Previous Brazilian studies described an incidence of 2.15 cases/10,000 live births in São Paulo, [26] 3 cases/10.000 live births in Rio de Janeiro, and 2.69 cases/10.000 live births in Rio Grande do Sul, all in the vicinity of our 2.87 cases/10,000 live births.

The mean maternal age in GS cases was 20.8 years and it was 27.3 among non-GS mothers, and there was an association between young maternal age and GS (p<0.01).

The association between GS and young maternal age has been frequently documented. [9, 25–27, 29, 30] In South Brazil, less than 2% of mothers (n=19) were \geq 35 years old, with an incidence of 0,31 cases/10,000 live births in this age group. When only mothers younger than 19 years are considered, the incidence reaches its highest number, with 8.83 cases/10,000 live births.

GS was associated with fewer years of maternal formal education (p<0.01), single parenting (p<0.01), and a smaller number of prenatal consultations (p<0.01).

No association was found between single/multiple pregnancies and the presence of GS (p=0.46).

There was an association between GS and birth <37 weeks (p<0.01). Anderson et al. [27] found no difference in the length of prenatal care between patients with and without GS. In South Brazil, women pregnant with babies with GS had a significantly smaller number of prenatal consultations, which is worrisome given the complexity of this malformation and the high risk of preterm delivery. [27, 30] Egger et al. [29] found a 5.7 relative risk of premature birth for GS. Indeed, 53.3% of babies with this malformation in South Brazil (613/1150, Table 2) were born <37 weeks of gestation, and there was an association between preterm delivery and GS.

Although cesarean section delivery is not beneficial in GS, [26, 30–32] most babies with the malformation (1016/1224, 83%, p<0.01) were delivered via cesarean section, and an association between the malformation and mode of delivery was found. Perhaps obstetricians and mothers feel more comfortable with a surgical delivery in the setting of malformations.

The mean BW for GS babies was 2341.2 g, while for non-GS babies was 3189.4 g, and there was an association between GS and lower BW (p<0.01).

The median Apgar 1 and Apgar 5 in GS cases was 8 and 9, respectively, and in non-GS babies was 9 and 10. This difference was statistically significant (p<0.01).

GS is associated with low and very low BW, [27, 30] and we found an association between weight <2500 g and GS. Furthermore, neonates with GS presented lower Apgar scores than neonates without the anomaly.

Analysis by sex (p=0.10) showed no statistically significant difference, as previously reported. [24,26, 29]

Although no difference was found between GS and non-GS babies regarding race (p=0.96), international studies reported differences in prevalence concerning race/color. [27, 30]

Limitations: Because this study was based on secondary data, the ascertainment of information was conditional on the completeness and accuracy of the available records and the level of clinical detail obtained was limited. Stillbirths were not included due to an error when generating data, which was reported to the website, but the problem was not solved. Despite these limitations, this large population-based study estimates the prevalence and demographic factors associated with CDH, EA, and GS, and extends the limited descriptive epidemiologic information available.

CONCLUSION

This study provides valuable information regarding three important malformations that carry an important impact on affected patients and families. The incidence of CDH in South Brazil is 0.93 cases per 10,000 live births, and there is an association between CDH and preterm birth, cesarean delivery, lower BW, lower Apgar, male sex, and black ethnicity. EA incidence is 0.47 cases per 10,000 live births, and an association between the malformation and mothers ≥35 years, multiple pregnancies, premature birth, cesarean delivery, lower BW, and Apgar was found. GS incidence is 2.87 cases per 10,000 live births. There is an association between the malformation and young maternal age, fewer years of maternal formal education, single parenting, a smaller number of prenatal consultations, premature birth, cesarean delivery, lower BW, and lower Apgar.

This study adds to the limited information on these congenital anomalies in Latin America and shows regional differences regarding the association of all three malformations and race when compared to publications from other countries.

Acknowledgements: Nil
Conflict of Interest: None.
Source of Support: Nil

Consent to Publication: Author(s) declared taking informed written consent for the publication of clinical photographs/material (if any used), from the legal guardian of the patient with an understanding that every effort will be made to conceal the identity of the patient, however it cannot be guaranteed

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

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