

Epidemiological Analysis of Osteomuscular Anomalies in Southwest Maranhão

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Análisis Epidemiológico de Anomalías Osteomusculares en el Sudoeste de Maranhão

RESUMO

As anomalias congênitas osteomusculares são alterações morfológicas, que ocorrem durante o desenvolvimento embrionário ou fetal, repercutindo na ausência total e/ou parcial de membros superiores e/ou inferiores. **Objetivo:** analisar os casos de anomalias congênitas osteomusculares no sudoeste do Maranhão. **Método:** Trata-se de estudo ecológico, de séries-temporais, realizado com dados sobre nascidos vivos da Unidade Gestora Regional de Saúde de Imperatriz (2009-2018). **Resultados:** As anomalias osteomusculares representaram 43% de todas as anomalias notificadas, com maior prevalência em Porto Franco, Sítio Novo, Amarante do Maranhão, Imperatriz e Estreito. Predominaram gestantes com menos de 35 anos, sem parceiro, pretas/pardas, com gravidez única e parto vaginal. Entre os recém-nascidos, houve maior frequência de crianças com peso adequado, nascidas a termo e do sexo masculino. **Conclusão:** Evidencia-se que essas anomalias representam um agravamento de saúde pública, que pode trazer complicações aos acometidos, logo, é essencial o diagnóstico oportuno e seguimento dos casos.

DESCRIPTORES: Anomalias Congênitas Osteomusculares; Epidemiologia; Maranhão

ABSTRACT

Congenital musculoskeletal anomalies are morphological changes that occur during embryonic or fetal development, resulting in the total and/or partial absence of upper and/or lower limbs. **Objective:** to analyze cases of congenital musculoskeletal anomalies in southwestern Maranhão. **Method:** This is an ecological, time-series study, carried out with data on live births from the Imperatriz Regional Health Management Unit (2009-2018). **Results:** Musculoskeletal anomalies represented 43% of all reported anomalies, with a higher prevalence in Porto Franco, Sítio Novo, Amarante do Maranhão, Imperatriz and Estreito. There was a predominance of pregnant women under 35 years of age, without a partner, black/brown, with a single pregnancy and vaginal birth. Among newborns, there was a greater frequency of children with adequate weight, born at term and male. **Conclusion:** It is clear that these anomalies represent a public health problem, which can cause complications for those affected, therefore, timely diagnosis and follow-up of cases are essential.

DESCRIPTORS: Congenital Musculoskeletal Anomalies; Epidemiology; Maranhão.

RESUMEN

Las anomalías musculoesqueléticas congénitas son cambios morfológicos que ocurren durante el desarrollo embrionario o fetal, dando como resultado la ausencia total y/o parcial de los miembros superiores y/o inferiores. **Objetivo:** analizar casos de anomalías musculoesqueléticas congénitas en el suroeste de Maranhão. **Método:** Se trata de un estudio ecológico, de series de tiempo, realizado con datos de nacidos vivos de la Unidad de Gestión Regional de Salud de Imperatriz (2009-2018). **Resultados:** Las anomalías musculoesqueléticas representaron el 43% de todas las anomalías reportadas, con mayor prevalencia en Porto Franco, Sítio Novo, Amarante do Maranhão, Imperatriz y Estreito. Hubo predominio de gestantes menores de 35 años, sin pareja, de color negro/pardo, con embarazo único y parto vaginal. Entre los recién nacidos hubo mayor frecuencia de niños con peso adecuado, nacidos a término y del sexo masculino. **Conclusión:** Es claro que estas anomalías representan un problema de salud pública, que puede causar complicaciones a los afectados, por lo que el diagnóstico oportuno y el seguimiento de los casos son fundamentales.

DESCRIPTORES: Anomalías musculoesqueléticas congénitas; Epidemiología; Maranhão.

RECEIVED: 12/08/2025 APPROVED: 12/26/2025

How to cite this article: Furtado JAC, Silva HL, Santos ARS, Ramos WA, Dias ICCM. Epidemiological Analysis of Osteomuscular Anomalies in Southwest Maranhão. Saúde Coletiva (Brazilian Edition) [Internet]. 2026 [cited year month day];17(104):18890-18905. Available from: DOI: 10.36489/saudecoletiva.2026v17i104p18890-18905

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INTRODUCTION

Congenital musculoskeletal anomalies (CMA) result from morphological changes that occur during embryonic or fetal development and may be apparent at birth. They represent an important cause of disability and infant mortality, caused by genetic, environmental, or multifactorial factors and, in many cases, with unknown etiology⁽¹⁾.

Worldwide, approximately 2% to 5% of live births (LB) have some type of CA. In Brazil, in 2017, they represented the second leading cause of death in children under 5 years of age, with an estimated 5% occurrence among the number of LBs⁽²⁾.

It should be noted that among the most frequent CA are malformations of the musculoskeletal system ICD-10 (Q65-Q79), followed by those of the nervous system⁽³⁻⁴⁾. A study conducted in Recife, Brazil, showed that musculoskeletal CA accounted for 44% of CA cases in 2013 and 2014⁽⁵⁾.

A subclassification of musculoskeletal CA shows that limb defects are the most frequent among them. The prevalence at birth varies according to the type of deformity, with polydactyly and congenital foot deformities being the most common. Muscu-

loskeletal malformations can affect newborns in different ways, including total and/or partial absence of limbs, as well as involvement of the upper and lower extremities, hands, and feet⁽⁶⁾.

It should be noted that maternal variables can contribute to the incidence of CA, such as age, number of pregnancies, type of delivery, number of prenatal consultations, race/color, level of education, and pre-existing and/or acquired diseases during pregnancy. In addition, some factors, mainly related to lifestyle, such as exposure to teratogenic substances during pregnancy and multiple sexual partners, may favor its occurrence⁽¹⁾.

It should be noted that there are preventive measures that can reduce the risks of CA, including family planning, with the aim of reducing extreme maternal age, guidance on the use of medications, as well as combating teratogenic substances during pregnancy⁽⁷⁾. These preventive measures can be addressed during prenatal consultations, as they provide an opportunity to monitor pregnancy, detect possible risks for CA, and provide knowledge and autonomy for self-care.

Considering the importance of knowledge about maternal and neo-

natal characteristics, as well as information about pregnancy and childbirth, the Brazilian Ministry of Health implemented the Live Birth Information System (SINASC) in 1990, with the aim of providing a national database capable of supporting the registration and monitoring of live births. The system is based on the Declaration of Live Births (DNV), where all information on LBs must be recorded⁽⁹⁾. However, it was only in 1999 that a specific field for recording AC was included in the DNV, making SINASC an important tool for monitoring these conditions⁽⁵⁾.

It is noteworthy that in 2014, a study was conducted that analyzed cases of AC in São Luís, Maranhão, Brazil, from 2002 to 2011, and showed an increase in the number of AC cases, from 0.34% in 2002 to 0.75% in 2011⁽¹⁰⁾. Another study showed that from 2006 to 2016, ACs were responsible for 2.96% of fetal deaths in the state of Maranhão⁽¹¹⁾.

There is a need for studies that analyze data on CA in other geographical areas of the state, given the disparity in the Human Development Index (HDI) between municipalities, cultural diversity, and the plurality of the healthcare network, which is concentrated mainly in larger cities such

as Imperatriz.

In view of the above, the objective was to analyze cases of congenital musculoskeletal anomalies in southwestern Maranhão.

METHOD

This is an ecological, time-series study with a quantitative approach⁽¹²⁾ that was conducted using live birth data from the Regional Health Department of Imperatriz, Maranhão, Brazil, from 2009 to 2018, also available in SINASC.

The regional health departments were created as a result of the decentralization of the Unified Health System (SUS) in order to provide comprehensive health care at all levels of care, serving as important tools for coordination and agreement between municipalities⁽¹³⁾. The state of Maranhão has 19 regional health authorities, one of which is the Imperatriz Regional Health Management Unit (UGRSI), which comprises 16 municipalities.

The city of Imperatriz has more than 270,000 inhabitants, making it the second largest city in Maranhão. It is located 632 kilometers from the capital São Luís⁽¹⁵⁾ and represents the UGRSI, which serves approximately 496,891 inhabitants⁽¹⁴⁾.

The data were obtained through the Health Surveillance Service (SVS) of the UGRSI. All newborns diagnosed with musculoskeletal CA, born between January 1, 2009, and December 31, 2018, residing in the municipalities that comprise the UGRSI, were included. Data from newborns with errors in the completion or incompleteness of information in the maternal and neonatal variables were excluded, with the information "ignored" not considered as incomplete information.

The presence or absence of musculoskeletal CA was established as the dependent variable. The independent

variables were divided into maternal and neonatal variables, with the maternal variables being: age, education, marital status, race/color, number of prenatal consultations, number of pregnancies, type of pregnancy, and type of delivery. The neonatal variables were: birth weight (low weight: < 2,499 g, ideal weight: 2,500 g to 3,999 g, overweight: ≥ 4,000 g), race/color, sex, classification of the newborn (NB) as preterm (up to 36 weeks and 6 days), term (37 weeks to 41 weeks and 6 days), and post-term (≥ 42 weeks), according to the parame-

$$x = \frac{n^{\circ} \text{ de NV com AC osteomusculares}}{n^{\circ} \text{ de nascidos vivos}} \times 10.000$$

In addition, the prevalence of musculoskeletal CA by municipality in the health region was calculated. *Microsoft Excel software*, version 2021, was used to tabulate the data.

With regard to ethical aspects, the study complies with Resolution 510, dated April 7, 2016, of the National Health Council. This study is derived from the research project entitled "Congenital anomalies in southwestern Maranhão: profile, spatial distribution, professional knowledge, and family experiences," approved by the

ters used by the Brazilian Ministry of Health⁽¹⁶⁾.

The selected variables were analyzed and tabulated descriptively, and the mean, absolute frequency (Fa), and relative frequency (Fr%) were calculated. The prevalence of musculoskeletal CA among all live births in the same period (X) and the prevalence of births with musculoskeletal CA among the number of live births with some type of congenital anomaly in the same period (Y) were calculated, as recommended by the Brazilian Ministry of Health.

$$y = \frac{n^{\circ} \text{ de NV com AC osteomusculares}}{n^{\circ} \text{ de nascidos vivos com AC}} \times 10.000$$

Research Ethics Committee of the Federal University of Maranhão, under opinion number 3,593,095.

RESULTS

Between 2009 and 2018, there were 96,354 live births in the UGRSI, of which 600 (0.62%) had CA, and 258 (43.00%) were musculoskeletal. Table 1 shows the numbers and prevalence of musculoskeletal CA per year of occurrence.

Table 1 – Number of NVs with CA and prevalence of musculoskeletal CA per year of occurrence in the Regional Health Department of Imperatriz, Maranhão, Brazil (2009-2018).

Year	NW with CA	Musculoskeletal	Musculoskeletal
Prevalence 104 Hab.			
2009	80	37	4,62
2010	64	35	5,46
2011	63	24	3,80
2012	54	29	5,37
2013	40	12	3,00
2014	54	24	4,44
2015	79	29	3,67
2016	74	26	3,51
2017	67	31	4,62
2018	23	11	4,78
Ignorado	2	0	0,00
TOTAL	600	258	4,30

Source: UGRSI/SINASC. Adapted by the authors (2019).

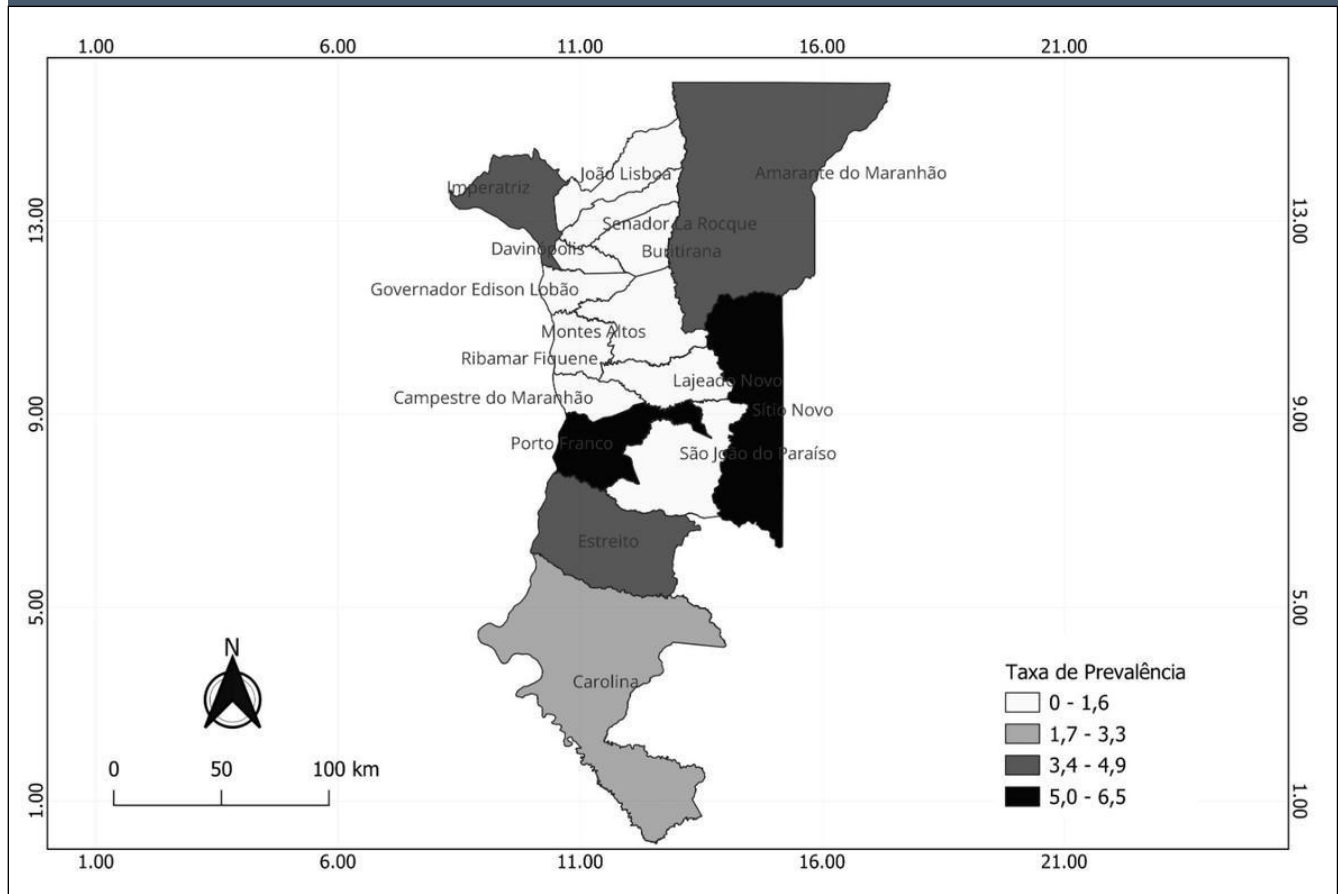
It can be observed that the highest prevalence of musculoskeletal WMSDs occurred in 2012 (5.37:10,000), while the lowest was

in the following year, 2013 (3.00:10,000).

Regarding the occurrence of musculoskeletal ACs in the municipalities of

UGRSI, Figure 1 shows the prevalence and an analysis of the regions with the highest occurrence.

Figure 1 – Prevalence of musculoskeletal ACs by municipalities in the Imperatriz Regional Health District, Maranhão, Brazil (2009-2018).



Source: UGRSI/SINASC. Adapted by the authors (2019).

The municipalities of Porto Franco (6.50:10,000) and Sítio Novo (5:10,000) stood out with the high-

est prevalence of musculoskeletal ACs. Next were Amarante do Maranhão (4.66:10,000), Imperatriz (4.14:10,000), and Estreito (3.40:10,000). The municipality of

Carolina had the lowest prevalence rate among the municipalities of the UGRSI in the period analyzed (2.85:10,000).

Table 2 presents the data regarding

Tabela 2 – Características maternas e neonatais dos casos de AC na Regional de Saúde de Imperatriz, Maranhão, Brasil (2009-2018).

Variables	General CA (n=600)		Musculoskeletal (n=258)		Variable	General CA (n=600)		Musculoskeletal (n=258)	
	Fa	Fr%	Fa	Fr		Fa	Fr%	Fa	Fr%
Maternal variables									
Age					Type of delivery				
<35 years	547	91.16	246	95.34	Cesarean	311	51.83	118	45.73
≥35 years	53	8.83	12	4.65	Vaginal	289	48.16	140	54.26

Quantitative Article

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Education					Type of Pregnancy				
Elementary School	49	8.16	26	10.07	Single	584	97.33	255	98.83%
High school	184	30.66	71	27.51	Double	14	2.33	2	0.77
Higher education	366	61.00	160	62.01	Triple	1	0.16%	1	0.38
Ignored	1	0.16	1	0.38	Ignored	1	0.16	0	0.00
Marital status					No. of Pregnancies				
With partner	144	24	66	25.58	Primipara	0	0.00%	0	0
No partner	454	75.66	191	74.03	Multipara	590	98.33	257	99.61
Ignored	2	0.33	1	0.38	Ignored	10	1.66	1	0.38
Race/Color					Prenatal consultations				
Black or brown	385	64.16	160	62.01	Up to 6 consultations	317	52.83	129	50.00
White	4	6.66	18	6.97	> 6 consultations	0	0	0	0
Yellow or Indigenous	11	1.83	2	0.77	Ignored	283	47.16	129	50.00
Ignored	164	27.33	78	30.23					
Neonatal variables									
Birth weight (grams)					Gestational age (weeks)				
< 2500	134	22.33	41	15.89	Up to 36 weeks and 6 days	89	14.83	34	13.17
2500 to 3999	430	71.66	203	78.68	37s to 41s6d	321	53.50	140	54.26
≥ 4000	35	5.83	14	5.42	≥ 42s	19	3.16	6	2.32%
Ignored	1	0.16	0	0.00	Ignored	171	28.50	78	30.23
Race/color					Gender				
Brown	143	23.83	70	27.13	Female	245	40.83	113	43.79
Indigenous	3	0.50	0	0.00	Male	350	58.33	145	56.20
White	15	2.50	7	2.71	Ignored	0	0.00	0	0.00
Ignored	439	73.16	181	70.15					

Source: UGRSI/SINASC. Adapted by the authors (2019).

Musculoskeletal CA were more prevalent in newborns whose mothers were younger than 35 years old (95.34%), had attended or were attending higher education (62.01%), without a partner (74.03%), black or brown (62.01%), multiparous (99.61%), with a single fetus (98.83%), and who had a vaginal delivery (54.26%). Regarding prenatal consultations, 50.00% of the records had the field ignored and 50.00% had up to 6 consultations.

Regarding neonatal musculoskeletal variables, newborns with ideal weight (78.68%), male sex (56.20%), and full-term (54.26%) predominated. The race/color field was ignored in

70.15% of the records.

DISCUSSION

In Brazil, musculoskeletal CA are most frequent in the southeast and northeast regions⁽³⁾. The high prevalence of cases in the southeast region may be related to its population size, as well as greater activity in CA registration in SINASC⁽³⁾. In the northeast region, the high number of cases may be related to the rate of consanguinity, the use of teratogenic substances, medications such as misoprostol and thalidomide, and the possible low intake of folic acid by women of reproductive age⁽⁴⁾.

In this sense, the findings of this

study are similar to studies conducted in other states in the northeast region of the country. In Alagoas, the prevalence of musculoskeletal CA was 44.8%⁽⁸⁾ and in Bahia, 48.04%⁽¹⁷⁾. In addition, there was similarity in a study that analyzed the epidemiological scenario of the capital of Maranhão, pointing to a prevalence of 48%⁽¹⁰⁾.

It should be noted that musculoskeletal CA are more easily reported because they are easily visible in the immediate postpartum period or even on ultrasound scans during prenatal care. This allows for diagnosis at the time of completing the DNV shortly after birth, unlike other CA, such as congenital heart disease, which re-

quire more robust tests for diagnosis⁽⁴⁾.

Another factor is the high prevalence of congenital foot deformities, classified by the International Classification of Diseases (ICD-10) as Q66, representing the main malformation of this system, reaching 13,516 cases in Brazil between 2013 and 2017, which is equivalent to 29% of musculoskeletal CA⁽³⁾.

The study also analyzed the prevalence of musculoskeletal CA according to the municipalities that make up the UGRSI. It is noted that neighboring municipalities had different prevalences, while others had no recorded cases, similar to the findings of a study⁽⁴⁾, which also surveyed cases in Maranhão and found differences between municipalities geographically close to the capital.

Geographical differences in prevalence can be explained by population, environmental, genetic, and/or social differences. They may also be related to cases in which the diagnosis was made after discharge from the maternity ward and completion of the DNV, in addition to being influenced by the manner in which the information was collected, and need to be investigated more specifically in future studies⁽⁸⁾. Findings such as these are relevant because they can be used to identify populations with demographic characteristics that favor the presence of malformations and/or identify specific environmental teratogens⁽¹⁸⁾.

Regarding maternal age, the findings are similar to those in the scientific literature. In a study⁽⁸⁾ conducted in Alagoas, Brazil, 75.5% of mothers were under 35 years of age. In a study conducted in Recife, Brazil, 49.01% of mothers were between 20 and 29 years old. Extreme ages are variables that are related to the development of malformations and are considered a biological risk factor during pregnancy.

It should be noted that women over 35 years of age may have pregnancies with an increased risk of chromosomal

malformations, aneuploidies that are associated with a decrease and aging of the eggs. Meanwhile, young mothers may have an increased risk of musculoskeletal and nervous system malformations, as well as those caused by external factors, such as the misuse of abortifacient drugs and alcohol consumption, with the use of psychoactive substances hindering access to and attention to health care during pregnancy⁽¹⁹⁻²⁰⁻²¹⁾.

With regard to education, the prevalence of higher education in this study is inconsistent with the scientific literature on risk factors for CA. Low educational attainment is a risk factor for pregnancy, since women with incomplete schooling have more difficulty accessing prenatal care and important information during pregnancy, as well as difficulties in understanding the risks of gestational diseases, nutritional information, supplementation, adequate weight, among others^(8,20). This finding raises questions about possible factors that may override education in the risk of CA, such as extreme age, multiparity, or marital status.

In this context, women who do not have marital stability are at greater risk for CA, since this situation brings psychological insecurity and lower family income, in addition to reproductive risk with the involvement of multiple sexual partners and exposure to sexually transmitted infections, such as syphilis⁽²²⁾.

Another important point is that skin color is also a determining factor in the number of children. Black and brown women have larger families⁽²³⁾. In southeastern Brazil, white women have the lowest fertility rate in the country (1.55 children per woman), while in northern Brazil, black women have more children (2.67 children per woman)⁽²²⁾. This may be related to the occurrence of CA, given that the greater the number of pregnancies, the greater the chance of exposure to risk factors for CA.

In this sense, prenatal care is essential in the prevention of CA, since one of its main goals is to identify extrinsic factors that may contribute to the onset of CA⁽⁸⁾. The study found that half of the pregnant women had up to six prenatal consultations, which is fewer than the number recommended by the Brazilian Ministry of Health, which establishes six or more consultations. Another finding is the number of "ignored" cases, which requires attention, given the importance of prenatal care.

The number of consultations may not correlate with the existence of CA, however, a reduction in consultations may favor an increase in risk factors for its development, since prenatal care is an important tool for prevention and guidance regarding exposure to environmental and teratogenic factors^(8,24).

Regarding the choice of delivery type, the findings differ from the literature, where cesarean delivery is most frequent in the presence of CA. This data is linked to the choice of surgical delivery in the face of a previous diagnosis of malformation and the possibility of surgical intervention in the neonatal period. In musculoskeletal CA, surgical delivery is often indicated due to the impossibility of passing through the vaginal canal⁽²⁰⁻²¹⁾. Even so, more than half of the pregnant women in this study had vaginal delivery, raising questions about early identification in prenatal care, including those highlighted by the low number of consultations.

With regard to neonatal variables, low birth weight is generally related to the presence of CA, as well as being one of the main causes of infant mortality, newborn weakness, and a precursor to delayed child development⁽¹⁹⁾. However, the study showed a higher frequency of musculoskeletal CA among those born with adequate weight. This finding may be related to variables that were not investigated, such as maternal overweight and

pre-pregnancy diseases.

Regarding the race/color of newborns, the study found a large number of "unknowns" (70.15%), which hinders research on this variable. However, among the records that had this field filled in, there was a predominance of brown skin color (27.13%), a finding similar to that of a study conducted in Alagoas, Brazil⁽⁸⁾.

Regarding the sex of the newborns, the survey showed a predominance of males among those affected by musculoskeletal CA (56.20%), evidence also found in Brazilian studies conducted in Rio Grande do Sul, Minas Gerais, and Alagoas^(8,22,25).

Some studies have pointed to the presence of CA as one of the main factors for prematurity^(19,20,24). However, in the present study, a predominance of full-term NV (54.26%) was observed, similar to that found in an-

other study conducted in the capital of Maranhão⁽²⁶⁾.

It should be noted that the reliability and quality of the data entered in the DNV depends on the correct completion by the responsible professionals, since there is a possibility of errors during completion. The importance of training and periodic data evaluations in search of improvements within the system and identification of possible failures is highlighted.

CONCLUSION

Musculoskeletal CA were prevalent among the others, and there was a difference in prevalence between the municipalities that make up the Imperatriz health region. The cities with the highest prevalence were Porto Franco, Sítio Novo, Amarante do Maranhão, Imperatriz, and Estreito.

The cases predominated in young mothers, without partners, black/brown, with single pregnancies and vaginal deliveries. In relation to newborns, children with ideal weight, born at term and male.

Further research is needed to identify other variables in the groups analyzed and to promote maternal and child health actions for the prevention, early diagnosis, and management of CA, as well as the qualifications of the professionals responsible for completing the DNV, as it is the instrument from which investigations can be carried out.

It should be noted that the findings of this investigation corroborate similar studies conducted in the state capital and in Brazil, which confirms the importance of musculoskeletal CA as a public health problem.

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