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RESEARCH

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POST-SURGICAL DEATHS IN NEONATES WITH CONGENITAL MALFORMATIONS IN THE DIGESTIVE OR MUSCULOSKELETAL SYSTEMS

Óbitos pós-cirúrgicos em neonatos com malformações congênitas do aparelho digestivo ou osteomuscular

Muertes post-quirúrgicas en neonatos con malformaciones congénitas digestivas o musculoesqueléticas

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ABSTRACT

Objective: To analyze the occurrence of post-surgical deaths in newborns with malformation in the digestive or musculoskeletal systems in a reference maternity hospital. **Method:** This is an exploratory and retrospective study, with a quantitative approach, conducted in a reference maternity located in Teresina – PI. Data were collected from Tabwin and infant death investigation forms of neonates born in 2016 and 2017 and analyzed through the Statistical Package for the Social Sciences software. **Results:** The most prevalent type of malformation of the digestive and musculoskeletal systems among neonates who died after surgery was the imperforate anus (41%) and gastroschisis (64.2%), respectively. Septic shock, followed by acute kidney failure, constituted the determining factors of the analyzed deaths. **Conclusion:** Early diagnosis is the key factor for reducing morbidity and mortality in neonates affected by congenital malformations, as it contributes to the targeting and planning of care actions essential for these patients.

DESCRIPTORS: Infant, Newborn; Congenital abnormalities; Abdominal wall; Digestive system; Infant mortality.

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RESUMO

Objetivo: Analisar a ocorrência de óbitos pós-cirúrgicos em recém-nascidos com malformação do aparelho digestivo ou osteomuscular em uma maternidade de referência. **Método:** Estudo exploratório, retrospectivo, de abordagem quantitativa, realizado em uma maternidade de referência localizada em Teresina – PI. Os dados foram coletados do *Tabwin* e de fichas de investigação de óbito infantil de neonatos nascidos em 2016 e 2017 e analisados no software *Statistical Package for the Social Sciences*. **Resultados:** O tipo de malformação mais prevalente do aparelho digestivo e osteomuscular entre os neonatos que foram a óbito após cirurgia foi o ânus imperfurado (41%) e a gastrosquise (64,2%), respectivamente. O choque séptico, seguido da insuficiência renal aguda foram os fatores determinantes dos óbitos analisados. **Conclusão:** O diagnóstico precoce é o fator primordial para redução da morbimortalidade de neonatos acometidos por malformações congênitas, uma vez que contribui para o direcionamento e planejamento dos cuidados imprescindíveis a esses pacientes.

DESCRITORES: Recém-nascido; Anormalidades congênitas; Parede abdominal; Sistema digestório; Mortalidade infantil.

RESUMEN

Objetivo: Analizar la ocurrencia de muertes post-quirúrgicas en recién nacidos con malformación digestiva o musculoesquelética en una maternidad de referencia. **Método:** Estudio exploratorio, retrospectivo, con enfoque cuantitativo, realizado en una maternidad de referencia ubicada en Teresina - PI. Los datos se recopilaron de *Tabwin* y de registros de investigación de muerte infantil de neonatos en 2016 y 2017 y se analizaron utilizando el programa *Statistical Package for the Social Sciences*. **Resultados:** El tipo de malformación digestiva y musculoesquelética más frecuente entre los neonatos que murieron después de la cirugía fue el ano imperforado (41%) y la gastrosquisis (64,2%), respectivamente. El shock séptico, seguido de insuficiencia renal aguda, constituyeron los factores determinantes de las muertes analizadas. **Conclusión:** El diagnóstico temprano es el factor principal para reducir la morbimortalidad en los neonatos afectados por malformaciones congénitas, ya que contribuye a la dirección y planificación de la atención esencial para estos pacientes.

DESCRIPTORES: Recién nacido; Anomalías congénitas; Pared abdominal; Sistema digestivo; Mortalidad infantil.

INTRODUCTION

The assessment of infant mortality in a country is carried out using the Infant Mortality Rate (IMT), since this indicator allows the assessment of the levels of social and economic development of a country, and also allows the evaluation of the health conditions of a given population. In Brazil, the main causes of infant death are related to prematurity, infections and congenital malformations, the latter being the most prevalent in the south, southeast and central-west region and the white population the most affected.[1-2]

Congenital malformations constitute any functional or morphological alteration identified before birth or shortly after delivery, which may be of environmental or genetic cause, and are divided between those that do not cause significant damage to the carrier, the so-called minor malformations, while the largest malformations are those that need surgical intervention and usually leave important permanent side-effects for the person affected. The major congenital malformations that affect the digestive and musculoskeletal systems stand out in relation to the other systems, since they currently occur markedly.[3-4]

The newborn (NB) affected by congenital malformation has a high risk of progressing to death, and its survival is associated with factors such as the type and severity of the anomaly, the clinical characteristics of the patient and access to appropriate surgical treatment. Prematurity and low socioeconomic status are also variables that interfere with their survival. Thus, the late referral of newborns with congenital malformations for surgical correction is a contributing factor to the increase in neonatal mortality rates.[4-5]

Considering that patients at extreme ages, such as newborns, have an increased risk of evolving to death when undergoing surgical procedures, due to the fragility of their systems, it is believed that the hospital's experience with the procedure, the resources available by the institution, the skill of the surgical team and pre and postoperative care are resources that can predict the clinical outcome of these patients.[6]

In view of the above and the fact that there is a shortage of national studies on post-surgical deaths in neonates with congenital malformations, the following was listed as a research problem: what is the occurrence of deaths in newborns with congenital malformation of the device or musculoskeletal system submitted to surgical procedures? Therefore, the objective of the study is: to analyze the occurrence of post-surgical deaths in newborns with malformations of the digestive or musculoskeletal system in a reference maternity hospital.

METHODS

Retrospective, exploratory and quantitative study carried out at a reference maternity in high obstetric and perinatal complexity care located in Teresina-Piauí. Only those newborns with digestive or musculoskeletal malformations who died in 2016 and 2017 were included in the study. Among newborns with musculoskeletal malformations, those with gastroschisis and omphalocele were specifically included. Neonates who died during surgery, had minor congenital anomaly, or even had their death attested before performing the surgical procedure were excluded from the study, thus leaving a total of 36 newborns, who died after performing the surgery to correct the malformation.

To collect the information, a form previously prepared by the authors of this study was used, containing clinical data on the newborn regarding congenital malformation, as well as the determinants of death. The data were collected from the infant death investigation forms provided by the Epidemiology Center of that institution, also obtaining the number of post-surgical deaths of neonates with digestive or musculoskeletal malformations. Tabwin version 3.0, a program used to tabulate data available on DATASUS, was also used as a data source, from which the total number of deaths from congenital malformations of the digestive and musculoskeletal system was consulted.

The data were organized and encoded in the *Microsoft® Excel® Software*, being subsequently imported and analyzed using the *Statistical Package for Social Sciences* (*SPSS®*) version

22.0. In addition, descriptive statistical analysis was performed using relative and absolute frequencies, as well as inferential analysis using the chi-square test with correction of *Fisher*'s exact test. In the analysis performed, a 95% confidence interval and a significance level of 5% ($p \leq 0.05$) were adopted. The results were arranged in tables and the discussion was based on the literature.

The study was carried out in accordance with the ethical and legal precepts of CNS Resolution 466/2012, and its data was collected after approval by the Research Ethics Committee of the State University of Piauí - UESPI, on October 3, 2018, under opinion number 2.936.586 and CAAE number 97621918.5.0000.5209.

RESULTS AND DISCUSSION

During the period from January 2016 to December 2017, 188 newborns died in the studied maternity, whose cause of death was the presence of some type of congenital malformation of the digestive or musculoskeletal system. Among these, 36 post-surgical deaths were identified, of which 14 (38.9%) had a congenital malformation of the musculoskeletal system (gastroschisis or omphalocele), and 22 (61.1%) some malformation of the digestive system.

Data referring to the clinical profile of newborns with congenital malformations of the digestive or musculoskeletal system who died after surgery are presented in table 1 and show that among those of the digestive system there was a predominance of males 12 (54, 5%), pre-term newborns 12 (54.5%), with low birth weight 12 (54.5%) and who had a cesarean section 14 (63.6%) as the prevailing type of delivery. Most of these newborns 9 (40.9%) had a survival of seven days or more.

In relation to the clinical profile of newborns with musculoskeletal congenital malformations who died after surgery, the male gender 8 (57.1%) was predominant, and the full-term newborns 8 (57.1%) had a higher representativeness. Low birth weight was seen in 9 (64.3%) newborns and most deliveries were cesarean type 10 (71.4%). Most of these newborns 6 (42.9%) had a 28-day or longer survival rate.

Table 1 - Clinical profile of newborns with congenital malformations of the digestive or musculoskeletal system who died after surgery. Teresina, PI, Brazil, 2018 (N = 36).

Variables	Digestive System		Musculoskeletal System	
	N (22)	%	N (14)	%
Sex				
Female	10	45.5	6	42.9
Male	12	54.5	8	57.1
Gestational age				
Pre-term (≥36w)	12	54.4	6	42.9
Full-term (≥37w)	10	45.5	8	57.1
Birth weight				
Low weight (<2500g)	12	54.5	9	64.3
Suitable weight (≥2500g)	10	45.5	5	35.7
Type of delivery				
Vaginal	8	36.4	4	28.6
Cesarean	14	63.6	10	71.4
Survival Rate				
< a week	5	22.7	3	21.4
≥ a week	9	40.9	5	35.7
≥ 28 days	8	36.4	6	42.9

*The total sample may vary due to the inadequate filling of the infant death investigation forms regarding the performance of a surgical procedure.

It was observed that among the predominant types of malformations of the digestive system, nine (41%) newborns with imperforated anus and five (22.8%) with esophageal atresia without fistula stood out. With regard to the musculoskeletal malformations studied, there was a higher incidence of newborns affected by gastroschisis 9 (64.2%), as shown in Table 2.

Table 2 - Most prevalent malformations of the digestive and musculoskeletal systems among newborns who died after surgery. Teresina, PI, Brazil (N = 36).

Type of malformation	N	%
Digestive System	22	100
Esophageal atresia without fistula	5	22.8
Esophageal atresia with fistula	2	9
Duodenal atresia	1	4.5
Duodenal stenosis	2	9
Megacolon	3	13.7
Imperforated anus	9	41
Musculoskeletal System	**14**	**100**
Omphalocele	5	35.8
Gastroschisis	9	64.2

It was also found that among the newborns studied, 11 (30.5%) had septic shock as the determining cause of death. Acute renal failure (AKI) was also identified as an important cause, being present in 4 (11.1%) of newborns with congenital digestive or musculoskeletal malformations. These and other findings are shown in table 3.

Table 3 - Evaluation of the determinants related to post-surgical death of newborns with congenital digestive or musculoskeletal malformations. Teresina, PI, Brazil, 2018 (N = 36).

Causes of deaths		
Determining factor	**N**	**%**
Septic shock	11	30.5
Cadiogenic shock	2	5.6
Hemorrhagic syndrome	1	2.8
Pulmonary hemorrhage	3	8.3
TGI hemorrhage	3	8.3
Sepsis	3	8.3
Hypovolemic shock	3	8.3
Cardiorespiratory arrest	2	5.6
Disseminated intravascular coagulation (DIC)	2	5.6
Respiratory failure	2	5.6
AKI	4	11.1

Table 4 shows the association between the variables: type of congenital malformation found in the study and the weight of the newborn affected by such malformation, and in this it was found that all types of malformations have significant statistical correlation (p = 0.019). The occurrence of omphalocele is strongly associated with newborns with low birth weight. Also noteworthy is the presence of esophageal atresia without fistula in very low birth weight infants, gastroschisis in extremely low birth weight infants and imperforated anus in children born with adequate weight.

Table 4 - Association between congenital malformation and birth weight of newborns affected by congenital malformation of the digestive or musculoskeletal system who died after surgery. Teresina, PI, Brazil, 2018 (N = 36).

Types of malformation	Birth weight				P-value
	Extremely low (<1000g)	Very low (<1500g)	Low (<2500g)	Adequate (2500 - 4000g)	
	n (%)	n (%)	n (%)	n (%)	
Esophageal atresia without fistula	3 (21.4)	2 (33.2)	0 (0.0)	0 (0.0)	0.019[1]
Esophageal atresia with fistula	1 (7.1)	1 (16.7)	0 (0.0)	0 (0.0)	0.019
Duodenal atresia	0 (0.0)	1 (16.7)	0 (0.0)	0 (0.0)	0.019
Duodenal stenosis	0 (0.0)	0 (0.0)	0 (0.0)	2 (13.3)	0.019
Megacolon	1 (7.1)	1 (16.7)	0 (0.0)	1 (6.7)	0.019
Imperforated anus	2 (14.4)	0 (0.0)	0 (0.0)	7 (46.7)	0.019
Omphalocele	1 (7.1)	1 (16.7)	1 (100.0)	2 (13.3)	0.019
Gastroschisis	6 (42.9)	0 (0.0)	0 (0.0)	3 (20.0)	0.019

[1] *Fisher*'s exact test

Regarding the population investigated in this study, there was a predominance of males, both in malformations of the digestive system and in those of the musculoskeletal system. This finding corroborates with another study where the male gender was also present in the majority (55.9%) of newborns who had some congenital malformation.[7]

Much is discussed about the greater occurrence of anomalies among live male births, and studies increasingly point out that mainly chromosomal anomalies are by nature more frequent, thus not excluding other malformations such as those related to the digestive and musculoskeletal systems.[8]

It was found in this study that birth with gestational age less than or equal to 36 weeks prevailed among cases of malformations of the digestive system, therefore, they were newborns considered premature. In contrast to this finding, a study showed that newborns with digestive malformations were predominantly born full-term, representing 57.7%.[9]

Regarding the musculoskeletal system, the prevalence of neonates born full-term was found, which corroborates the finding of a study in which it was found that 75.5% of newborns affected by malformation were born full-term, that is, 37 weeks or more.[10] On the other hand, another study showed that neonates with abdominal wall defects, such as gastroschisis and omphalocele, have a higher chance of premature delivery.[11] It was also shown that prematurity contributes to death rates related to gastroschisis, since it is considered as one of the most recurrent complications associated with this malformation.[12]

When directing the discussion to the relationship between gestational age and the presence of malformation, the literature points to a higher frequency of malformation among neonates born prematurely when compared to those with gestational age of 37 weeks or more. The association between the birth defect and birth is not yet well defined, so it is not known whether the presence of a malformation can be a causative factor in shorter duration of pregnancy.[8]

These circumstances may occur in cases where the identification of the congenital anomaly still occurs in intrauterine life and there is an interruption of pregnancy so that interventions can be performed in the framework of the anomaly, or in cases where the anomaly itself is the factor that compromises the development of the fetus, thus triggering premature birth.[13]

It was evident in this study that the analysis of the variable birth weight was similar between the categories of malformations studied, which was more prevalent in neonates born with low weight. These data differ from the findings in the literature, where adequate birth weight was found to be predominant among neonates who had some type of digestive malformation or abdominal wall defects.[9,14]

Therefore, it is understood that the birth of a newborn weighing between three and four kilos is essential, because it favors a better adaptation to extrauterine life, since the infant mortality rates in this weight range are lower than those with low birth weight.[8]

Regarding the type of delivery, the predominance of the cesarean type was found among the studied malformations. Several studies have shown that two out of three live births with congenital malformations are born by caesarean section. It is considered a much higher rate when compared to newborns who do not have malformation.[8]

This type of delivery is most often the most used, since the determination of cesarean deliveries is related to factors such as pre-existing morbidities and early detection of anomaly.[15] This can be explained by the high rates of newborns affected by malformations that are still diagnosed in prenatal care, which reflects in the choice of scheduled delivery.[14]

When analyzing the prevalence of congenital malformations of the digestive system among the population studied, we found a greater occurrence of imperforated anus, a finding that differs from that found in the literature, where it was evidenced that esophageal atresia and colon atresia or stenosis are the most prevalent digestive malformations. This finding can be explained by the fact that congenital malformations are closely related to the social and epidemiological context in which they are inserted, showing themselves in a varied way according to the region studied.[14,16]

There was a prevalence of gastroschisis among malformations of the musculoskeletal system, this finding corroborates with that found in the literature, which evidenced its predominance among musculoskeletal malformations. Among the most evident anomalies are those related to musculoskeletal deformities, followed by those of the central nervous system, genitourinary system and chromosomal anomalies.[4,13]

Their predominance among the others is due to the fact that they are identified at birth and, in some cases, even identified during prenatal care, during exams such as morphological ultrasound and nuchal translucency measurement, enabling a diagnosis more accurate postpartum.[13]

The determining factor for the deaths of NBs with malformations submitted to surgical procedures in this study was the same found in the national literature, which points out that septic shock is the main cause of death among newborns admitted to critical care units, representing a total of 40.83%.[17]

Thus, the use of specific antibiotics, the presence of a hospital infection control team working in health services, availability of minimally invasive surgical techniques and technological resources, in addition to multidisciplinary care in neonatal intensive care units can contribute to the increase in survival of these newborns.[10,13,15]

It is also worth mentioning that, with regard to the association between birth weight and the studied congenital malformations, there was a statistically significant association. Numerous studies report that the association and the rationale are generally based on the relationship with prematurity and the delay in fetal development due to the anomaly in question. Low birth weight appears to be associated with congenital heart disease and musculoskeletal defects in some encephalopathies.[9,13-14]

CONCLUSION

The perforated anus and gastroschisis were the most common malformations among neonates who died after surgery at the maternity in the study. The characteristics of newborns affected by digestive malformations are similar to those affected by gastroschisis and omphalocele (musculoskeletal malformations), with a predominance of low birth weight NBs, born by cesarean section, male, and with a hospital stay longer than one week until evolving to death. The variable gestational age diverged between the two categories of malformations studied, where those related to the digestive system were predominant in premature newborns, and in musculoskeletal ones, full-term neonates had a higher occurrence.

It was also found that post-surgical deaths were mostly caused by septic shock and acute renal failure. It is believed that in order to have a longer survival of these newborns, early diagnosis is essential, since it can contribute to the direction and planning of appropriate care for the sick newborn, thus reducing their morbidity and mortality.

The results found may in turn not reliably represent the investigated population, since the incomplete and / or inadequate filling of the infant death investigation forms, regarding the record of the surgical procedure to correct the malformation, was identified as an important limitation of this study. Such information started to be filled in satisfactorily only in the year 2017, which may have led to a reduction in the number of newborns included in this study.

However, it is hoped that the results of this study may contribute to awaken the multidisciplinary team that provides direct assistance to these NBs, a reflection on their actions, since one of the main determining factors for death in the studied maternity is an avoidable cause.

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