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ORIGINAL ARTICLE

Positional cranial deformities in preterm infants and their association with health indicators

Deformidades craneales posicionales en lactantes prematuros y asociación con indicadores de salud

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What do we know about the subject matter of this study?

Preterm infants are at increased risk for positional skull deformities due to the malleable cranial characteristics and long periods of hospitalization in neonatal intensive care units.

What does this study contribute to what is already known?

We analyzed the association between gestational age, hospitalization time, and presence of positional skull deformities in children with a history of prematurity attending an Early Care Program (ECP), observing mild positional plagiocephaly in most of the children, independent of the degree of prematurity. It is suggested that a cephalometric evaluation should be performed on every child who enters an ECP.

Abstract

Prematurity is a risk factor for positional cranial deformities since preterm infants have a more malleable skull and are susceptible to deformities due to external pressures. **Objectives:** To describe positional cranial deformities and peri/postnatal pathologies in preterm infants and to analyze the association between gestational age, birth weight, length of hospitalization, and severity of cranial deformities measured by the Cranial Vault Asymmetry Index (CVAI) and the Cephalic Index (CI). **Patients and Method:** Analytic, cross-sectional study. 103 preterm infants aged under 4 months of corrected age admitted during 2017 to an Early Intervention Program (EIP) were included. Participants were classified according to gestational age as follows: extremely preterm (< 28 weeks), very preterm (28-32 weeks), and moderate-to-late preterm (32-37 weeks). Head circumference, anteroposterior diameter, width, and head diagonals were measured, and the CVAI and CI were calculated. Peri- and

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postnatal history was obtained from clinical records. **Results:** 103 preterm infants were evaluated (17 extremely preterm, 78 very preterm, and 8 moderate-to-late preterm). 99 (96.1%) of the preterm infants had positional cranial deformity and, regardless of the degree of prematurity, presented similar cranial anthropometric measurements. Mild plagiocephaly was the most frequent cranial deformity in all groups. We observed a positive association between the days of hospitalization and the CVAI and there was no relationship between the degree of prematurity and the severity of the positional cranial deformation. **Conclusions:** Most of the patients admitted to the EIP presented positional cranial deformities, mainly mild plagiocephaly, regardless of the degree of prematurity. The presence of plagiocephaly was positively associated with prolonged periods of hospitalization. No relationship was confirmed between the degree of prematurity and the severity of the positional cranial deformity.

Introduction

Positional skull deformities are an alteration that affects the shape of the head due to external pressures on the skull, which can occur *in utero* or during the first months of life[1-3]. Risk factors include prematurity, long periods of hospitalization, male sex, delayed psychomotor development, congenital torticollis, multiple pregnancy, and instrumental delivery, among others[1,3-8]. Prematurity is particularly identified as one of the most important risk factors due to the malleable characteristic of the skull bones of this group of children, being more susceptible to cranial modeling, associated with a proportionally larger cephalic mass and less neck control[1,3].

Annually, 15 million premature babies are born in the world, being the first cause of perinatal morbidity in developed countries[9]. In Chile, 8.2% of newborns are born before 37 weeks of gestation, and 1.2% are premature at less than 32 weeks of gestational age[9]. According to the World Health Organization(WHO), any newborn born alive before 37 weeks of gestational age is considered preterm and is divided into the following categories: extremely preterm (< 28 weeks), very preterm (28 to 32 weeks), and moderate to late preterm (32 to 37 weeks)[10].

The probability of occurrence of positional skull deformities would be greater the higher the degree of prematurity[11]. In addition, prolonged hospitalization has been associated with the presence of positional skull deformities in infancy, defined as a hospital stay that varies in a wide range from 3 to more than 30 days, although there is still no consensus among researchers regarding the number of days[12]. Long periods of hospitalization at birth due to severe illness and mechanical ventilation have been strongly associated with the development of positional skull deformities, even in term infants[11].

Sometimes, the asymmetric growth of the skull is accompanied by facial asymmetry, which can affect the positioning of the eyes, ears, forehead, cheeks, and jaw[1,11,13,14]. Some authors even indicate that it could participate in the appearance of visual alterations such as astigmatism and strabismus due to a delay in the development of the visual field, as well as auditory alterations in cases of severe deformities[4,15]. In addition, it is postulated that children with positional skull deformities could be associated with developmental delays, such as gross motor development alterations[6], as well as language delay, learning difficulties, and attentional deficit[15], without determining a linear causal relationship.

Depending on the area where the preferential support of the head is generated, several types of positional deformities can occur such as plagiocephaly, which is characterized by a flattening in the parieto-occipital area; brachycephaly, where there is a bilateral occipital flattening, kind of symmetrical, without side preference; and dolichocephaly, where there is a flattening of the temporo-parietal region. Of the above, positional plagiocephaly is the most common type of skull deformity in infancy[1,3,16].

The approach should be early to avoid eventual secondary effects on the development of the craniofacial mass of the child[5,6, 9,11,15,17], recommending prevention, screening, and early treatment for better results, highlighting also that early intervention will imply lower associated costs[8,18].

The diagnosis is fundamentally clinical, and, to date, several instruments contribute to the diagnostic evaluation of this condition, such as anthropometric calipers, photographs, and software for digital analysis[5,6,17], which are scarce or nonexistent in many Chilean neonatal intensive care units.

Although the incidence of positional skull deformities is difficult to determine since it corresponds to an underdiagnosed condition, it is estimated that, internationally, it could affect up to 46-48% of children under one year of age[3,5,19]. In Chile, there are no epidemiological data available on the prevalence of these alterations either in the general population or in the population at risk.

The objectives of this study were to describe positional skull deformities and peri/postnatal pathologies in infants attending an Early Care Program (ECP) and to analyze the association between gestational age, birth weight, and hospitalization time with the severity of the skull deformity measured by the Cranial Vault Asymmetry Index (CVAI) and the Cephalometric Index (CI).

Patients and Method

Study design

Quantitative, non-experimental, analytical, cross-sectional study with convenience sampling.

Population and sample

Newborns referred from the Neonatology Follow-up Polyclinic of the *Hospital Dr. Gustavo Fricke* (HGF) of Viña del Mar to the ECP belonging to the School of Kinesiology of the *Pontificia Universidad Católica de Valparaíso* (PUCV), after hospital discharge check-up, in the context of the national follow-up of premature and high-risk children.

During 2017, 130 infants were referred to the ECP, whose objective is to prevent, identify, and treat possible alterations or delays in the psychomotor development of children with a high-risk history, with follow-ups and outpatient physiotherapy interventions during their first 3 years of life. Most of the children admitted to the ECP had a history of prematurity and other neurobiological risk factors such as genetic syndromes and perinatal hypoxia, among others. During their hospital stay in the neonatology unit of the HGF, they were frequently repositioned as a preventive measure for the development of positional skull deformities.

The children included in the study met the following criteria: prematurity, having an anthropometric head evaluation with a caliper on admission to the program, and having a corrected age ≤ 3 months and 29 days at the time of the evaluation, thus obtaining a sample of 103 infants born prematurely. The study was approved by the Bioethics Committee of the *Pontificia Universidad Católica de Valparaíso* (BIOE-PUCV-H150-2017) and the parents and/or primary caregivers of the participants signed an informed consent before entering the study.

Variables and instruments

Health indicators at birth

Gestational age, birth weight, and weight for gestational age according to intrauterine growth curves by Alarcón and Pittaluga[20] were considered. The infants were classified into three groups according to their gestational age according to WHO criteria: extremely preterm (< 28 weeks), very preterm (28 to 32 weeks), and moderate to late preterm (32 to 37 weeks)[10].

Time of hospitalization

Length of hospital stay is considered prolonged when it is longer than 10 days, according to the cut-off point of reference established by Gold et al in 2016[21].

Measurement age

Corrected age measured in days at which the anthropometric evaluations were performed.

Head anthropometry measurements

Head circumference (in cm), length, width, and major and minor diagonal of the skull (in mm) were measured[22].

Cephalometric Index (CI)

Allows for numerically assessing the presence and degree of dolichocephaly or brachycephaly considering as a reference the parameters published by Bosch and Costa in 2017[22]. Brachycephaly is considered as mild (CI = 86-90), moderate (CI = 91-100), and severe (CI > 100); dolichocephaly as mild (CI = 70-74), moderate (CI = 60-69), severe (CI < 60).

Cranial Vault Asymmetry Index (CVAI)

Evaluates the presence and degree of plagiocephaly considering as a reference the parameters published by Bosch and Costa in 2017, where the higher the score the greater the severity of the deformity[22]. Mild plagiocephaly is considered as 0-10 mm, moderate as 10-20 mm, and as severe > 20 mm.

Peri- and postnatal pathologies

Jaundice of prematurity, anemia of prematurity, hyaline membrane disease, bronchopulmonary dysplasia, hypoxia, sepsis, intrauterine growth restriction, patent ductus arteriosus, intracranial hemorrhage grade I, II, III, and IV, periventricular leukomalacia, hypocalcemia, other metabolic disorders, chromosomopathy, and torticollis were considered.

Statistical analysis

The analysis of results was performed with IBM-SPSS Statistics® version 25. The Shapiro-Wilk test was used for the variable analysis to measure normal distribution. The description of the qualitative variables was presented as frequency and percentage. Quantitative variables were presented as mean and standard deviation when normal distribution was observed and as median and interquartile range when a non-normal distribution was observed. To determine the differences in quantitative variables between participants according to the degree of prematurity, the one-way ANOVA or Kruskal-Wallis test was used depending on the normal distribution of the variables.

To evaluate the association between categorical variables, Fisher's exact test was used and to evaluate the association between risk factors and CVAI, Pearson's correlation coefficient or Spearman's correlation coefficient was used depending on the normal distribution of the variables. A significance level of $\alpha = 0.05$ was considered and p-values < 0.05 were considered significant differences.

Results

Initially, 130 infants were evaluated and 27 participants were excluded, 1 due to a history of pathologies that generate a skull deformation other than positional, 5 did not have the signature of informed consent by the parents and/or caregivers, and 21 did not meet the corrected age criteria for anthropometric measurements. The final sample consisted of 103 infants (Moderate to late preterm n = 8, Very preterm n = 78, Extremely preterm n = 17). At birth, the infants had a median gestational age of 29.9 ±2.5 weeks and a mean weight of 1,403.0 ± 446.7 grams. At the time of the anthropometric head measurement, the infants had a median age of 30.0 ± 24.9 days of corrected age, and the following nutritional status: 8.9% were undernourished, 13.9% were at risk of undernutrition, 53.2% presented normal weight, 21.5% were overweight, and 2.5% were obese (according to anthropometric W/A and W/L curves)[23]

Table 1 presents birth health indicators, head anthropometric measurements, corrected age at the time of measurement, and severity of the skull deformation according to the degree of prematurity. As expected, it was observed that extremely preterm infants presented significantly lower birth weight ($p < 0.0001$) than very preterm and moderate to late preterm infants. In addition, it was observed that extremely preterm infants spent significantly more time in the hospital ($p < 0.0001$) than very preterm and moderate to late preterm infants. Furthermore, no significant differences were observed in the corrected age at the time of the anthropometric head assessment of the infants. The infants, regardless of the degree of prematurity, presented similar circumference, length, and width of the skull. Also, it was observed that the CVAI and CI were similar in the three groups.

Regarding skull deformities, 99 infants (96.1%) had some positional skull deformity. There was no association between the degree of prematurity and the severity of positional skull deformity. When described by the type of skull deformity, a high percentage of infants with plagiocephaly was observed, with mild plagiocephaly being the most frequent in all degrees of prematurity. A low percentage of infants, regardless of the degree of prematurity, presented brachycephaly and dolichocephaly (Table 1).

Table 2 shows the peri and postnatal pathologies according to the degree of prematurity. It was observed that there is an association between the degree of prematurity and the presence of peri- and postnatal pathologies. Based on the results of Fisher's exact test, it can be stated that extremely preterm infants present more jaundice of prematurity, anemia of prematurity, hyaline membrane disease, sepsis, intrauterine growth restriction, bronchopulmonary dysplasia, intracranial hemorrhage grade I and II, and patent ductus arteriosus compared with very preterm and moderate to late preterm infants. There were no cases of torticollis or chromosomopathies.

The association between risk factors for positional skull deformity was analyzed, finding a positive and significant correlation between days of hospital stay and CVAI in all infants (Rho = 0.237; $p = 0.015$), suggesting that the longer the infants were hospitalized, the higher the CVAI, an indicator of plagiocephaly (Table 3) (Figure 1).

Discussion

In this study, it was observed that the infants, regardless of the degree of prematurity, presented similar head anthropometric measurements. Only 3.9% had no positional skull deformity in the group studied. There was a high percentage of infants with positional plagiocephaly (94.2%), with mild plagiocephaly being the most frequent in all degrees of prematurity. There was no relationship between the degree of prematurity and the severity of positional skull deformity, but there was a positive association between days of hospitalization and CVAI in all the infants studied. One possible explanation for this is that preventive measures during hospitalization may have been applied more rigorously in the most premature infants, in the context of possible insufficient human resources and prioritizing according to risk.

On the other hand, extremely preterm infants present more anemia of prematurity, hyaline membrane disease, sepsis, bronchopulmonary dysplasia, and intracranial hemorrhage grade I and II compared with very preterm and moderate to late preterm infants. It should be noted that, although a higher degree of prematurity is associated with a greater presence of pathologies, in this study, there was no association between the presence of these pathologies and positional skull deformity.

Table 1. Birth health indicators, head anthropometric measurements, corrected age at the time of measurement

	Moderate to late preterm (n: 8)	Very preterm (n: 78)	Extremely preterm (n: 17)		
Birth health indicators	M/Me ± SD/inter-quartile range	M/Me ± SD/inter-quartile range	M/Me ± SD/inter-quartile range		ANOVA or Kruskal-Walis p value
Birth weight (gr)	1614.4 ± 392.9 a	1490.3 ± 416.9 a	903.1 ± 206.4 b		< 0.0001
Length of hospitalization (days)	32.1 ± 10.3 a	48.2 ± 22.3 a	103.4 ± 17.7 b		< 0.0001
Measurement age (corrected age/days)	52.0 [10.3-54]	30.0 [19-55.8]	29.0 [21.5-38]		0.785
Birth weight category	N (%)	N (%)	N (%)		Fisher's exact test p value
Adequate weight for gestational age	3 (37.5%)	43 (55.1%)	13 (76.5%)		
Small for gestational age	2 (25%)	14 (17.9%)	0 (0%)		
Severe small for gestational age	3 (37.5%)	15 (19.2%)	2 (11.8%)		0.320
Large for gestational age	0 (0%)	6 (7.7%)	2 (11.8%)		
Head anthropometry measurements	M/Me ± SD	M/Me ± SD	M/Me ± SD		ANOVA p value
Head circumference (in cm),	37.1 ± 1.6	37.9 ± 2.0	37.3 ± 2.0		0.332
Skull length (in mm)	127.8 ± 5.5	127.9 ± 7.0	126.5 ± 7.3		0.758
Skull width (in mm)	100.8 ± 3.3	103.7 ± 6.5	103.3 ± 7.0		0.463
Major diagonal (in mm)	124.1 ± 5.0	126.8 ± 6.5	125.3 ± 6.6		0.405
Minor diagonal (in mm)	119.8 ± 4.7	121.7 ± 6.7	120.6 ± 6.1		0.640
Cranial Vault Asymmetry Index (CVAI)	4.4 ± 4.9	5.2 ± 4.5	4.7 ± 2.9		0.838
Cephalometric Index (CI)	79.0 ± 3.4	81.2 ± 4.7	81.9 ± 6.7		0.402
Positional skull deformation	N(%)	N(%)	N(%)	Total	Fisher's exact test p value
Without positional skull deformity	1 (12.5%)	3 (3.8%)	0 (0%)	4 (3.9%)	0.320
With positional skull deformity	7 (87.5%)	75 (96.2%)	17 (100%)	99 (96.1%)	
With one positional cranial deformity	6 (75%)	54 (69.2%)	9 (52.9%)	69 (67%)	
With two positional cranial deformities	1 (12.5%)	21 (26.9%)	8 (41.1%)	30 (29.1%)	0.232
With three positional cranial deformities	0 (0%)	0 (0%)	0 (0%)	0 (0%)	
Plagiocephaly	7 (87.5%)	74 (94.2%)	16 (94.2%)	97 (94.2%)	
Mild	5 (62.5%)	60 (76.9%)	14 (82.4%)	79 (76.7%)	
Moderate	2 (25%)	14 (17.9%)	2 (11.8%)	18 (17.5%)	0.651
Severe	0 (0%)	0 (0%)	0 (0%)	0 (0%)	
Brachycephaly	0 (0%)	13 (12.7%)	6 (35.3%)	19 (18.4%)	
Mild	0 (0%)	12 (15.4%)	5 (29.4%)	17 (16.5%)	
Moderate	0 (0%)	1 (1.3%)	1 (5.9%)	2 (1.9%)	0.200
Severe	0 (0%)	0 (0%)	0 (0%)	0 (0%)	
Dolichocephaly	1 (12.5%)	9 (11.6%)	3 (17.3%)	13 (12.8%)	
Mild	1 (12.5%)	8 (10.3%)	2 (11.8%)	11 (10.7%)	
Moderate	0 (0%)	1 (1.3%)	1 (5.9%)	2 (1.9%)	0.398
Severe	0 (0%)	0 (0%)	0 (0%)	0 (0%)	

The description of the qualitative variables was presented as frequency and percentage. Quantitative variables were presented as mean and standard deviation when normal distribution was observed and as median and interquartile range when a non-normal distribution was observed. p-value < 0.05 were considered significant differences.

Table 2. Peri- and postnatal pathologies of the sample

	Moderate to late preterm (n: 8) n (%)	Very preterm (n: 78) n (%)	Extremely preterm (n: 17) n (%)	Fisher's exact test p value
Neurological Conditions				
Intracranial hemorrhage grade I y II	0 (0%)	9 (11.5%)	9 (52.9%)	< 0.0001
Intracranial hemorrhage grade III y IV	0 (0%)	2 (2.6%)	2 (11.8%)	0.084
Hypoxia	1 (12.5%)	5 (6.4%)	0 (0%)	0.188
Periventricular leukomalacia	0 (0%)	8 (10.3%)	1 (5.9%)	0.878
Respiratory Conditions				
Bronchopulmonary dysplasia	2 (25%)	10 (12.8%)	11 (64.7%)	< 0.0001
Hyaline membrane disease	2 (25%)	44 (56.4%)	16 (94.1%)	0.000
Metabolic conditions				
Hypocalcemia	2 (25%)	23 (29.5%)	9 (52.9%)	0.083
Other metabolic disorders	0 (0%)	4 (5.1%)	3 (17.6%)	0.055
Cardiac Conditions				
Patent ductus arteriosus	0 (0%)	7 (9%)	4 (23.5%)	0.047
Systemic Conditions				
Anemia of prematurity	1 (12.5%)	44 (56.4%)	17 (100%)	< 0.0001
Jaundice of prematurity	6 (75%)	73 (93.6%)	17 (100%)	0.036
Sepsis	1 (12.5%)	32 (41%)	16 (94.1%)	< 0.0001
Intrauterine growth restriction	4 (50%)	25 (32.1%)	2 (11.8%)	0.038

Regarding plagiocephaly, in each group according to the degree of prematurity at birth, most of the infants presented mild or moderate plagiocephaly. These data agree with other studies that mention that prematurity is one of the main risk factors for presenting positional skull deformities in children[1,3-8]. This should be considered a warning sign, given the possible associated alterations[24,25], which may require some type of treatment, implying an additional expense for families and the healthcare system[26,27].

Table 3. Association between gestational age, birth weight, length of hospitalization with CVAI in the studied group

	Cranial Vault Asymmetry Index (CVAI)	
Variables	Rho	P value
Gestational age (weeks)	-0.170	0.081
Birth weight (gr)	-0.175	0.073
Time of hospital stay (days)	0.237	0.015*

The association was made with Spearman's linear correlation coefficient. *p < 0.05.

In relation to other risk factors for the development of positional plagiocephaly, the literature highlights prolonged hospitalization[28], a factor that in this study is associated with greater severity of positional plagiocephaly. Regarding the latter, the average days of hospitalization of the 3 groups according to gestational age at birth are classified as prolonged hospitalization. This may influence both the possibilities of spontaneous mobility and the positioning measures adopted during the peri- and postnatal periods, favoring the appearance of positional skull deformities[15]. It should be noted that, although the neonatal intensive care unit included in its care plan the frequent change of position during hospitalization, it seems that this measure was not sufficient to prevent the appearance of positional skull deformities.

Among the positional skull deformities found in this population, 94.2% of the infants presented plagiocephaly, 18.4% brachycephaly, and 12.8% dolichocephaly. These values partly agree with those published in other studies, which show that the most frequent positional skull deformity in children is plagiocephaly, followed by brachycephaly and dolichocephaly[29].

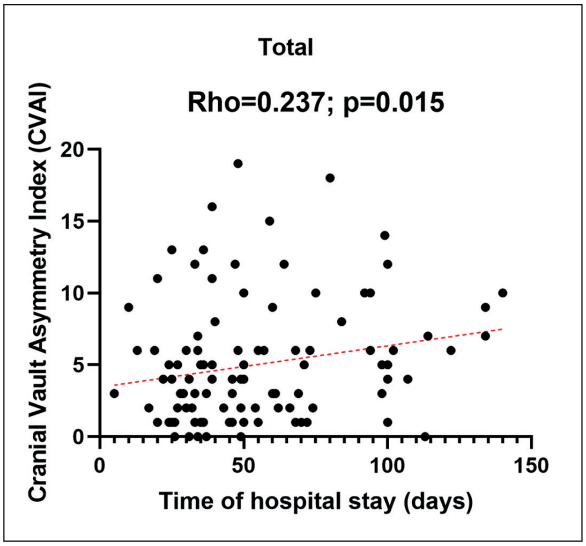

Figure 1. Association between days of hospital stay and CVAI. The association was made with Spearman's linear correlation coefficient. A significant association was considered $p < 0.05$.

Considering the high frequency of positional skull deformities present in this ECP and that anthropometric head measurements are a simple, easy, and low-cost evaluation[16], it would be important to include skull evaluation as a standardized and serial process for all infants admitted to a program like this, independent of the severity of prematurity, considering that most of the patients who attend neurodevelopmental follow-up plans present several risk factors for positional skull deformities such as prolonged hospitalization, possible mobility limitations, and delayed psychomotor development, among others[1,3-6,30].

It should be noted that no other national publications on the presence of positional skull deformities in premature infants attending early care programs were found in the reviewed literature. These results reinforce the need to establish early intervention programs, and even preventive ones, in neonatal and pediatric intensive care units, to avoid an increase in the incidence of cranial deformities and, especially, to avoid the appearance of deformities of greater severity, which would also result in lower associated health care costs.

This study has limitations that must be considered when interpreting the results. The population included belongs to only one health center and the sample is small and only represents what occurs in the ECP of the PUCV. It is suggested to replicate this study in Neonatology services of other cities in order to have provincial, regional, and national prevalence data of positional skull deformities and, therefore, to dimension this problem at a public health level.

On the other hand, in this study, only gestational age, birth weight, and time of hospitalization were associated with CVAI since the literature considers them among the main risk factors (11). The authors are aware that it was not possible to make an association with other risk factors in the analysis, such as the presence of health conditions associated with prematurity. It would be of great interest for a future study to focus on determining if there is an association between pre-, peri-, and post-natal pathologies and positional skull asymmetries in the Chilean population. In addition, the study design does not allow us to establish the causes of positional skull deformities, whether their presentation was of pre- or postnatal origin, nor the eventual impact on neurodevelopment. Future studies should further investigate the etiology and more effective interventions for the prevention and management of these alterations.

In conclusion, most of the preterm patients admitted to this ECP presented positional skull deformities, especially of the mild plagiocephaly type, independent of the degree of prematurity. CVAI was positively associated with prolonged hospitalization periods.

Intervention plans with a preventive character of these alterations should be implemented[31], both in neonatal and pediatric hospitalization units. In particular, the case of premature infants should be addressed who, due to their own condition and physiological hypotonia, have difficulty in changing their position independently during a large part of their hospital stay. Therefore, the appearance and magnitude of skull deformations will depend to a great extent on neonatal care[32], highlighting the protocols of frequent repositioning and the use of positioning devices, such as the preemie orthotic device[33] or other similar devices[34], whenever there is evidence of their usefulness. In addition, some authors consider that there may be an association between the presence of positional skull deformities and delayed psychomotor development[35].

All of the above reinforces the importance of paying special attention to this population[6,36,37], with the objective of timely screening and prevention of this condition[38]. Also, an early diagnosis should ideally be accompanied by an early start in the therapeutic process in the same service where the hospitalization occurred, in order to provide timely intervention and prevent the progression of the alteration.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

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