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Prevalence of depressive symptoms in patients with cleft lip and palate ☆,☆☆



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KEYWORDS

Cleft lip;
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Abstract

Introduction: Cleft lip and/or palate (CL/P) represent the most common congenital anomalies of the face.

Objective: To evaluate the prevalence of depressive symptoms in children and adolescents with nonsyndromic cleft lip and/or palate (nsCL/P).

Methods: We conducted an observational, case-control study, with a case study group composed of 61 patients with nsCL/P, aged 7–17 years, and a control group of 61 clinically normal patients. Both groups were selected at the same institution.

Results: Depressive symptoms were observed in the case group (nsCL/P), but there were no statistically significant differences compared to the control group. No association was found between the two groups (case and control) in relation to sociodemographic variables: gender, age and education.

Conclusions: This study identified the prevalence of depressive symptoms in children and adolescents with nsCL/P from a localized geographic population, although the results were not

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PALAVRAS-CHAVE

Fissura palatina;
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statistically significant when compared to the control group, not justifying the use of CDI (Child Depression Inventory) as a screening instrument for depressive symptoms in the examined population.

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Prevalência de sintomas depressivos em pacientes com fissuras labiopalatinas

Resumo

Introdução: Fissuras labiais e/ou palatinas (FL/Ps) representam as anomalias craniofaciais mais comuns.

Objetivos: Avaliar a prevalência de sintomas depressivos em crianças e adolescentes não sindrômicos com FL/P (FL/PNS).

Método: Foi realizado um estudo observacional de caso-controle com uma amostra populacional de conveniência, com um grupo caso (61 pacientes com FL/PNS, tendo idades entre 7 a 17 anos) e um grupo controle (61 pacientes clinicamente normais). Ambos os grupos foram selecionados na mesma Instituição.

Resultados: Sintomas depressivos foram observados no grupo caso (FL/PNS), mas não houve diferenças estatisticamente significantes quando comparado com o grupo controle. Não foi encontrada associação entre os dois grupos (caso e controle) em relação às variáveis sociodemográficas: gênero, idade e educação.

Conclusões: Este estudo observou a prevalência de sintomas depressivos em crianças e adolescentes com FL/PNS de uma população geográfica localizada, embora os resultados não tenham sido estatisticamente significantes quando comparado com o grupo controle, não justificando assim a utilização do Inventário de Depressão Infantil (IDI), como instrumento rastreador de sintomas depressivos na população analisada.

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Introduction

Nonsyndromic lip and/or palate cleft (NSCL/P) is the most prevalent congenital genetic defect of craniofacial area and results in anatomical complications and in psychological and behavioral disorders.¹ The incidence of NSCL/P varies according to geographic location, race and socioeconomic status,^{2,3} and has a mean distribution of about 1 case per 700 live births.⁴ The etiology of NSCL/P is multifactorial, involving several genes and complex molecular events that occur during embryogenesis, that are also influenced by environmental factors.^{5,6}

Some studies have suggested an increased risk for the development of psychiatric disorders in children and adolescents with NSCL/P, and have noted abnormal levels of depressive symptoms.⁷⁻¹² The literature contains population-based studies that identify depressive disorders in the general population, that reach a prevalence of 10% and an incidence of 2%.¹³ It is estimated that approximately 5% of people worldwide have depression, and about 10-25% of them may show some depressive episode during life.^{14,15}

The prevalence of depression in childhood increases with age, and is approximately 2% overall; it increases progressively and at adolescent ages, reaches levels close to

adulthood.¹⁶ In different regions of the world, including Brazil, these values range from 0.4 to 3.0% for children, and 3.3 to 12.4% for teenagers. These variations can be explained by methodological differences in sample selection strategy, as well as by cultural differences, depending on where the studies have been conducted.¹⁵⁻¹⁷

Early detection of depressive symptoms could indicate social, school or family environment damage,^{7,8,18} and, therefore, several methods are used for screening and diagnosis.^{19,20} The Child Depression Inventory (CDI) is used to assess depressive symptoms in children and adolescents in different clinical and research contexts.¹⁹⁻²¹ Thus, some studies have shown an association between occurrence of this malformation and psychosocial adjustment, suggesting greater attention to patients with NSCL/P, including their overall development and integration with the social environment.²¹⁻²⁴

In addition, psychiatric and psychological support of patients with NSCL/P throughout growth and development and also throughout the entire period of rehabilitation is necessary, in order to understand the needs of patients and of their parents who are feeling and living with a craniofacial malformation.^{10,11,23,24} Thus, the aim of this study was to assess the prevalence of depressive symptoms in children and adolescents with NSCL/P.

Methods

We conducted a case-control observational study. All participants were selected from the same institution (Centro de Referência para Reabilitação de Anomalias Craniofaciais e Clínicas Odontológicas e Médicas), in Minas Gerais, Brazil. The case study group consisted of 61 patients with NSCL/P,

all of them children and teenagers between the ages of 7 and 17; the control group consisted of 61 clinically healthy individuals (with negative personal or family history for craniofacial changes or syndromes), in the same case group. Both groups were selected by chance, enrolling an adequate number of subjects to equal the previous sampling calculations used to determine the number of subjects needed for

Table 1 Child Depression Inventory (CDI).

Data Collection Form – nº _____	
Date: ____/____/____	
1. Initials of the name:	Medical Record: _____
2. Gender:	1. Male ☐ ; 2. Female ☐ .
3. Date of Birth:	____/____/____ () years.
4. Nationality	4.1 Place of Birth: _____
5. Zone:	1. Urban ☐ ; 2. Rural ☐ .
6. Skin color:	1. Caucasian ☐ ; 2. African ancestry ☐ ; 3. Japanese ancestry ☐ ; 4. Indian ancestry ☐ ; 5. Other ☐ .
7. Degree of Instruction:	1. Illiterate ☐ ; 2. Elementary Incomplete ☐ ; 3. E. Elementary Complete ☐ ; 4. E. Secondary Incomplete ☐ ; 5. E. Secondary Complete ☐ ; 6. Not applicable ☐ .
8. Type of cleft:	1. PC (isolated) complete ☐ ; 2. PC (isolated) incomplete ☐ ; 3. LC (isolated) unilateral complete ☐ ; 4. LC (isolated) bilateral complete ☐ ; 5. LC (isolated) unilateral incomplete ☐ ; 6. LC bilateral incomplete ☐ ; 7. LC bilateral complete ☐ ; 8. LPC left unilateral complete ☐ ; 9. LPC right unilateral complete ☐ ; 10. LPC bilateral incomplete ☐ ; 11. LPC bilateral complete ☐ ; 12. Rare clefts (other) ☐ ; _____; 13. Not applicable ☐ .
This questionnaire consists of 27 groups of statements that describe your feelings and thoughts during the last two weeks . After reading carefully, choose the best statement (only one) in each group. Take care to read all the statements in each group before making your choice.	
1. () I get sad from time to time; () I am sad many times; () I am sad all the time	
2. () Nothing will ever work out for me; () I'm not sure if things will work out for me; () All will be fine for me	
3. () I do almost everything right; () Many things I do are wrong; () I do everything wrong	
4. () I have fun with many things; () I have fun with a few things; () I have no fun with anything	
5. () I am bad all the time; () Many times I am bad; () I am bad at one time or another	
6. () I think that bad things might happen to me one time or another; () I am worried that bad things would happen to me; () I am sure that terrible things will happen to me	
7. () I hate myself; () I do not like myself; () I like myself	
8. () All bad things happen by my fault; () Many bad things happen by my fault; () The bad things usually do not happen by my fault	

Table 1 (Continued)

9. () I do not think about killing myself; () I think about killing myself, but would not do it; () I want to kill myself
10. () I want to cry every day; () Many days I feel like crying; () I want to cry at one time or another
11. () Always there is something bothering me; () Often there is something bothering me; () Once in a while there's something bothering me
12. () I like being with other people; () I often do not like being with other people; () I have no desire to be with anyone
13. () I cannot decide on anything; () It is difficult to me to make decisions; () I decide myself about things easily
14. () My looks are cool; () There are some things I do not like about my appearance; () I am ugly
15. () I always have to force myself to do my homework; () I often have to force myself to do my homework; () I have no trouble doing schoolwork
16. () I have trouble sleeping every night; () I often have trouble sleeping; () I sleep well
17. () I get tired once in a while; () In many days I get tired; () I'm always tired
18. () On most days I'm not about to eat; () In many days I'm not about to eat; () I eat well
19. () I do not worry about pain; () I often worry about pain; () I always worry about pain
20. () I do not feel alone; () Many times I feel alone; () I always feel alone
21. () I never have fun at school; () I just have fun at school one time or another; () Many times I have fun at school
22. () I have many friends; () I have many friends, but want more; () I have no friends
23. () My performance in school is good; () My performance in school is not as good as before; () I am going wrong in matters on which I was good
24. () I will never be as good as others; () If I want I can be as good as others; () I am as good as others
25. () Nobody really loves me; () I am not sure if anybody loves me; () I am sure that somebody loves me
26. () I generally do what they tell me to do; () I generally do not do what they tell me to do; () I never do what they tell me to do
27. () I get along with people; () Often I get myself into fights; () I get myself into fights all the time
Sum (Total Score): _____

statistical purposes. Patients with CL/P as one manifestation of a clinical syndrome and those with a family history of consanguinity were excluded from the study. We compared the prevalence and severity of depressive symptoms between groups by applying CDI, a self-directed and self-explanatory questionnaire.¹⁹

This instrument (CDI) (Table 1) was developed in order to detect the presence and severity of depressive symptoms in children and adolescents so as to identify changes in mood, self worth, vegetative functions and self-evaluative and interpersonal behaviors. The 27 items of this questionnaire are summed to obtain a score that can be validated and specifically reflect the nationality of the questionnaire.^{19,25} During the application of the questionnaire, each item of CDI

was read in the presence of the patients to ensure understanding and remove uncertainties. The alpha value was set at 0.5%. A cut off point (score) of 17 points had been previously established for statistical significance. The results were compared using the chi-squared test and by logistic regression analysis. This study was approved by the Institutional Ethics Committee (Nº. 56/2010).

Results

The distribution of case ($n=61$) and control ($n=61$) groups followed a normal pattern for gender, and in both groups the most prevalent age was between 7 and 13 years (Table 2).

Table 2 General characteristics of patients with nonsyndromic lip and palate clefts (case group) and without lip and palate clefts (control group).

Variables	Cases		Controls		Total		OR adjusted	OR	CI 95%	<i>p</i>
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%				
<i>Gender</i>										
Male	29	47.5	27	44.3	56	45.9	1.14	1.13	0.56–2.32	0.716
Female	32	52.5	34	55.7	66	54.1				
<i>Age (years)</i>										
7–9	23	37.7	21	34.4	44	36.1	1.40	1.38	0.68–2.85	0.364
10–13	20	32.8	26	42.6	46	37.7				
14–17	18	29.5	14	23.0	32	26.2				
<i>Skin color</i>										
Leukoderma	35	57.4	24	39.3	59	48.3	2.07	2.0	1.00–4.27	0.048
Melanoderma	26	42.6	37	60.7	63	51.7				
<i>Depressive symptoms</i>										
Yes	13	21.3	08	13.1	21	17.2	1.80	1.77	0.68–4.70	0.234
No	48	78.7	53	86.9	101	82.8				
Total	61	100	61	100	122	100				

When the classification of NSCL/P was evaluated for the case group, we found a higher frequency of cleft palate ($n=33$), with equal distribution for isolated cleft lip and palate ($n=14$; corresponding to 23% for each in the group). When the two groups were compared, the logistic regression analysis identified no association among sociodemographic variables: gender ($p=0.716$; OR 1.14; CI 95% 0.56–2.32), age ($p=0.364$; OR 1.40; CI 95% 0.68–2.85) and schooling ($p=0.082$; OR 3.34; CI 95% 0.86–13.0). An association was detected for skin color ($p=0.048$; OR 3.34; CI 95% 1.0–4.27), meaning that individuals with NSCL/P are 3.34 times more likely to be Caucasians (Table 2).

We detected the presence of depressive symptoms in 21.3%, $n=13$ of the case group (NSCL/P); however, there was no statistical significance compared to the control group ($p=0.234$, OR 1.80, 95% CI 0.68–4.70) (Table 2). The analysis of each of the 27 items of CDI identified a homogeneous distribution for the frequency of depressive symptoms in both groups. An association for three items was observed, when we compared the mean of scores, by CDI sub-items (Table 3). Concerning the item related to suicide, it was observed that 8.4% ($n=5$) of subjects in the case group (nsL/PC) reported some suicidal thoughts compared to only 1.6% ($n=1$) in the control group.

Discussion

Some studies have reported an increased risk for the development of psychiatric disorders in children and adolescents with NSCL/P, citing abnormally high levels of depressive symptoms.^{10,11,26–28} A meta-analysis that included 340 studies identified high levels of depressive symptoms in several chronic diseases or conditions, in comparison with normal individuals.²⁸ Marked differences were found in chronic fatigue syndrome, fibromyalgia, migraine, epilepsy and in CL/P. The presence of these symptoms was more prevalent in female patients and in individuals living in developing

Table 3 Comparison of mean scores by sub-item of CDI (p -value, comparison of medians by Kruskal–Wallis test) of patients with nonsyndromic lip and palate cleft (case group) and control patients ($n=122$).

Sub-item	Cases	Controls	<i>p</i>
1	0.312	0.213	0.443
2	0.410	0.525	0.223
3	0.267	0.361	0.167
4	0.328	0.360	0.710
5	0.082	0.213	0.065
6	0.738	0.836	0.344
7	0.066	0.131	0.301
8	0.262	0.328	0.278
9	0.393	0.426	0.429
10	0.213	0.213	0.845
11	0.574	0.705	0.356
12	0.262	0.410	0.155
13	0.508	0.508	1.000
14	0.328	0.426	0.278
15	0.951	0.574	0.021 ^a
16	0.148	0.344	0.052
17	0.426	0.492	0.735
18	0.328	0.230	0.671
19	0.820	0.951	0.393
20	0.328	0.262	0.519
21	0.312	0.312	0.656
22	0.295	0.262	0.802
23	0.312	0.377	0.958
24	0.508	0.787	0.044 ^a
25	0.131	0.295	0.039 ^a
26	0.377	0.180	0.094
27	0.164	0.098	0.519

^a p -value < 0.05.

countries.²⁸ In the present study, despite the presence of depressive symptoms in the case group with NSCL/P (21.3%, $n=13$), no positive statistical significance was observed, when compared with control group ($p=0.234$; OR 1.80; CI 95% 0.68–4.70). A stratified analysis showed no association of depressive symptoms in both groups (case *versus* control) in the combination of variables, such as gender ($p=0.145$) and age ($p=0.165$).

It is known that the early detection of depressive symptoms is important for identifying in advance any ill-effects on the family, social and school environments^{7,8,26–28} so that the application of screening and diagnostic methods can guide the treatment of these patients.^{19,29} In an Irish case-control study, the authors compared the presence of functional, psychological disorders, such as anxiety and depression, and also behavioral disorders, among 160 children and adolescents with NSCL/P and in 113 normal individuals.³⁰ An important association of changes in behavior and depressive symptoms in the presence of NSCL/P ($p<0.001$) was observed. In our study, a score of 17 was used, based on the adaptation and standardization of this instrument in Brazil.²⁶ The analysis of each of the 27 items of CDI identified a homogeneous distribution of the frequency of depressive symptoms in both groups, thus with no positive association. In the Irish study, with respect to facial appearance and difficulty with speech, patients were more unhappy than controls, and high suicide rates were observed, as in our study, with 8.4% ($n=5$) of the respondents in the case group (NSCL/P), compared with 1.6% ($n=1$) in the control group.³⁰ Another study found a high incidence of psychological disorders and difficulties with social interaction in patients with CL/P.¹⁰

With respect to cleft type, we found higher rates of problems related to anxiety, depression, and learning and speech difficulties in children with isolated palate cleft, compared to children with both lip and palate cleft.³¹ In the present study, when comparing the presence of depressive symptoms among patients in the case group (NSCL/P) *versus* control group, we found similarities with those results, but with no positive statistical correlation. Other studies also point to an association between the occurrence of this malformation and psychosocial adjustment, suggesting greater attention to patients with NSCL/P, including their overall development and integration into the social environment.^{29,32–35} Although this study was conducted in a referral center for rehabilitation of craniofacial anomalies, it has some limitations, such as the limited geographic area, *i.e.*, restricted to a portion of a single Brazilian state. For all analyzes, alpha was 0.05. The sample size was sufficient for the detection of a difference of two points between groups in CDI instrument, considering a coefficient of variation of 0.5 and a statistical power of 0.8.

Although our results do not confirm the findings in the literature, this study suggests that psychological and psychiatric support is needed for patients with NSCL/P throughout their growth and development, and throughout the rehabilitation period, in order to understand the needs of patients and of their families who are experiencing first hand a craniofacial malformation.^{7,8,18,35} We also need to emphasize that in a limited geographic area and with the methodology and tool chosen, the use of a screening instrument for depressive symptoms for this group of individuals with

NSCL/P was not justifiable.^{10,11,23,24} Thus, the aim of this study was to assess the prevalence of depressive symptoms in children and adolescents with NSCL/P.

Conclusion

The authors of this study identified the prevalence of depressive symptoms in children and adolescents with nsCL/P from a localized geographic population, although the results were not statistically significant compared with the control group. There was no association of depressive symptoms with sociodemographic variables (gender, age and schooling). CDI is an instrument used in various chronic clinical and genetic conditions, aimed to identify patients with the potential or the risk of developing depressive symptoms. But in the specific population of the present study, we did not feel it necessary to adopt CDI as a screening instrument for such craniofacial abnormalities.

Conflicts of interest

The authors declare no conflicts of interest.

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