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RESEARCH ARTICLE

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PREVALENCE OF OROFACIAL PAIN IN ADULTS WITH CLEFT LIP/PALATE

^{*1}Gloria Maria Cortz Ravazzi, ²Dr. Rosane Sampaio Santos, ³Bianca Lopes Cavalcante de Leão, ⁴Dr. Rita de Cassia Tonocchi, ⁵Dr. Odilon Guariza-Filho and ⁶Dr. Jose Stechman-Neto

¹Dentistry; - Master's student Program in Communication Disorders, Tuiuti University of Paraná, Curitiba, PR, Brazil; ²Master's and Doctoral Program in Communication Disorders - Tuiuti University of Paraná, Curitiba, PR, Brazil; ³Doctor's student and Professor, Tuiuti University of Paraná, Curitiba, PR, Brazil; ⁴Master's and Doctoral Program in Communication Disorders - Tuiuti University of Paraná, Curitiba, PR, Brazil; Titular professor of Orthodontics in PUC-Pr., Curitiba, Pr, Brazil; ⁵Master's and Doctoral Program in Communication Disorders - Tuiuti University of Paraná, Curitiba, PR, Brazil

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*Corresponding author:

Gloria Maria Cortz Ravazzi

ABSTRACT

Objective: Functional disorders in individuals with cleft lip and palate (CL/P) can compromise structures of the stomatognathic system, which is one of the causes of orofacial pain in this group of patients. Numerous mechanisms of orofacial pain, particularly painful temporomandibular disorders (TMD-P), may be related to these conditions and have the potential to be the cause of diffuse. So, the objective of research was determine the prevalence of orofacial pain using validated instruments on individuals with CL/P. **Materials and Methods:** a observational study was conducted with an intentional random sample of patients with CL/P (n = 80). Two questionnaires were administered: a screening scale for painful TMD (AAOP) and the Graded Chronic Pain Scale (GCPS). **Results:** In the AAOP screening questionnaire, 75% of individuals with CL / P answered affirmatively to at least one question. The most significant questions were those related to noise (p = 0.007), mandibular movement (p = 0.032) and orofacial pain region (p = 0.044) demonstrated the importance of investigating TMD-P. Regarding the impact of chronic pain in relation to activities of daily living (GCPS), although no significant, it showed that for most individuals with CL / P, it has a low disability, regardless of its intensity (low / high), being more prevalent in older individuals (p = 0.013). **Conclusions:** the investigation of orofacial pain should be part of the routine at services that work with CL/P patients and should be counseled to seek help for such pain, which can affect their daily living.

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INTRODUCTION

Orofacial pain refers to any type of pain in the area bounded by the eyes and the lower mandibles, including the oral cavity. Exact prevalence data are lacking, although facial pain with or without accompanying symptoms appears to be a very frequent complaint [1]. The pain may radiate to different regions, such as the dental arches, ears, temples, forehead, occiput, and the cervical region of spine or shoulder girdle [2]. The aetiology of pain-related TMD is considered to be multifactorial and to result from a complex interaction between biological, psychological, social, and environmental variables [3, 4]. Functional disorders in individuals with cleft lip and palate (CL/P) can compromise structures of the stomatognathic system, which is one of the causes of complaints of orofacial pain in this population [5].

The different types of CL/P are closely linked to the orofacial muscles and structures adjacent to the stomatognathic system, which may be compromised in this condition [6]. Thus, CL/P may be associated with orofacial pain due to anatomic-functional connections. Some patients with CL/P have been submitted to numerous treatments over the years, such as plastic surgery, dental corrections and speech/hearing therapy, with the aim of normalizing both appearance (nose, lips and teeth) and function (speech, chewing, nasal breathing and hearing), but in this study it was found that general orofacial pain or psychosocial distress were no more common in this CL/P group than in a group without CL/P [7]. It is necessary to relate the complaints of patients to the current clinical history to ensure that all contributing factors, signs and symptoms, such as pain and difficulty performing jaw movements, are taken into consideration. It is also important to bear in mind that the diagnosis may be altered after the initial screening, as the assessment of chronic

conditions is an ongoing process. Moreover, the duration of the condition can be a determinant factor in the definition of the diagnosis, as acute and/or chronic pain can lead the clinician to a crossroads to determine the etiology of the disease[8]. Having instruments to assist in the diagnosis enables clinicians to guide patients to specialists, which, in turn, assists the patients in avoiding the inconvenience and time spent seeking adequate care. Thus, the patient history and preliminary physical examination should facilitate and guide the clinical investigation to a successful diagnosis and treatment [9]. Given the proximity of the structures, as demonstrated by Ferreira[10]there may be a relationship between the affected musculoskeletal regions and CL/P, since individuals with this condition can exhibit altered muscle anatomy, as stated by Sousa and Roncalli[11]. According to Liang[12],orofacial pain may be associated with mandibular asymmetry. Therefore, this disorder may be present in individuals with CL/P who have asymmetry due to the flawed formation of the structures of the stomatognathic system. Malocclusions, particularly of the transverse type where disrupted symmetry of the dental arches can be clinically observed, are a potential cause of functional disorders of the stomatognathic system. Hence, patientswith CLPs are potentially at risk of developing temporomandibular disorders (TMD), due to psychosocial burdens and malocclusions predisposing them to this condition [7, 13, 14]. The objectives of the present study were to estimate the prevalence of chronic orofacial pain in individuals with CL / P and to assess its impact as a function of pain-related intensity and disability with a brief measure of its severity in the assessed group.

MATERIAL AND METHODS

Ethical aspects: This study received approval from the human research ethics committee (certificate number 1.741.169) and was conducted in accordance with the guidelines stipulated in the Declaration of Helsinki.

Study design and Sample: This observational study was conducted with a sample selected consecutively of individuals with CL/P, selected in a one-month period. 209 subjects (n=209) with CL/P who appeared for routine evaluations at a reference center for facial malformations in southern Brazil were interviewed independently of the duration of treatment and type of cleft. Individuals less than 18 years of age, those with syndromes, those with comprehension difficulties and those unwilling to participate in the evaluations were excluded. The final sample was composed of 80 patients (n=80).

Data Collection: The data were collected by a single examiner who had undergone training and calibration exercises. A skilled, experienced professional in this specific field was considered the gold standard and trained the examiner in the use of the data collection instruments. Cohen's Kappa (K) index for intra-examiner (k = 0.92) and inter-examiner (k = 0.89) agreement revealed that the examiner was capable of collecting the data.

Collection Instruments (Questionnaires): - American Academy of Orofacial Pain (AAOP) screening questionnaire[9, 15]. The screening questionnaire proposed by the AAOP was administered to all individuals in both groups. This questionnaire is composed of ten questions with yes/no response options. Individuals with an affirmative answer to at least one of the questions were asked to answer the Graded Chronic Pain Scale. It should be stressed that the AAOP screening tool should not be the only justification for a more complex assessment, but rather an indication of the need for the administration of other assessment measures. Graded Chronic Pain Scale (GCPS)[16]has seven objective questions with response options scored from 0 (absence of pain) to 10 (worst possible pain). This scale was used to assess current pain, its extent and its interference with routine activities as well as the impact of pain on work and the capacity to control pain. This scale allows us to observe a hierarchical relationship between pain intensity and disability, in which the intensity of pain climbed the lower range of severity and the

disability climbed the upper range of gravity. The pain severity was graded into 4 steps:

- Grade I - low disability-low intensity;
- Grade II - low disability high intensity(no pain-related disability days in the prior 6 months);
- Grade III - high disability-moderately limiting (1-6 pain-related disability days);
- Grade IV - high disability-severely limiting (7 + disability days).

Statistical Analyses: The data were organized and submitted to statistical analyses using the SPSS (IBM Statistic 20.0®. Descriptive (frequency of variables) and bivariate (association tests and comparisons between groups) analyses were performed. The chi-squared test was used to test associations between categorical variables, with the level of significance set to 5%. For the purposes of analysis, CL/P was dichotomized as unilateral and bilateral, the GCPS score was categorized as “without pain in the previous six months”, “low disability” and “high disability”. The associations between the items on the AAOP questionnaire and GCPS with independent variable (CL/Puni or bilateral/gender) was evaluated using bivariate analysis. Nonparametric analyzes of the AAOP questionnaire score was performed in relation to the type of fissure and age and AAOP score in relation to the degree of chronic pain (GCPS) using the Kruskal-Wallis test.

RESULTS

Initially, 209 individuals were interviewed, of whom they were excluded by the exclusion criteria (individuals age <18years old; individuals with different syndromes; individuals who refused to participate). A total of 80 subjects met the inclusion criteria and answered the AAOP questionnaire. 53.8% (n = 43) were male and 46.3% (n = 37) were female, with an average age of 28.72 (± 11.42) years. Regarding the type of fissure, 58.8% (n = 47) were identified with unilateral fissure and 41.2% (n = 33) bilateral. In the sample evaluated, the scores of the screening questionnaire proposed by AAOP ranged from 0 to 6 points (0-10), and it can be said that the participants who presented pain of mild and moderate intensity (n = 80). Of the total number of individuals interviewed, 75% (n = 60) answered affirmatively at least one of the questions in the orofacial pain screening questionnaire (AAOP), and the chronic pain questionnaire (GCPS) was performed for this sample. The following are the positive responses to the AAOP screening questionnaire regarding the type of fissure and sex (Table 1). The total of affirmative responses from the AAOP screening questionnaire and the type of CL / P no statistically significant differences (p = 0.126). However, considering the questions in the AAOP questionnaire separately, the results were significant for question “Are you aware of noises in the jaw joints?”regarding the type of cleft (p = 0.007) with a relative risk of unilateral cleft compared to bilateral RR = 0.596 (0.398 - 0.891).Regarding the gender variable, there was a statistically significant difference in two questions. Question “Does your jaw get “stuck”, “locked” or “go out”?” (p = 0.032) with male relative risk in relation to female RR = 0.507 (0.405 - 0.633) and in question “Do you often have headaches, neck pain or dental pain?”(p = 0.044) also with male relative risk in relation to female RR = 0.602 (0.417 - 0.870).

The chronic pain questionnaire was applied to 60 individuals who answered affirmatively to one of the questions in the AAOP screening questionnaire. The answers were categorized into: no pain, Grade I and II (low disability-low intensity and low disability-high intensity), and Grade III and IV (high disability-moderately limiting and high disability-severely limiting). In the bivariate analyzes, there were no statistically significant differences in relation to the type of CL / P (p = 0.323) or to gender (p = 0.823). (Table 2). Table 3 shows the relationship between the degree of chronic pain and the average age, and the relationship between the AAOP and GCPS screening questionnaire.

Table 1. Absolute and percentage frequencies of positive answers to items on AAOP questionnaire in control and observational groups with odds ratios and respective 95% confidence intervals (n = 94)

	Control	CL/P	p-value	OR*	95% CI
Do you have difficulty, pain or both when opening your mouth, for instance, when yawning?	6 (21.4%)	6 (9.1%)	0.101	0.36	0.10 - 1.25
Does your jaw get "stuck", "locked" or "go out"?	1 (3.6%)	19 (28.8%)	0.006	10.91	1.38 - 86.13
Do you have difficulty, pain or both when chewing, speaking or using your jaws?	3 (10.7%)	16 (24.2%)	0.135	2.66	0.71 - 10.01
Are you aware of noises in the jaw joints?	12 (42.9%)	34 (51.5%)	0.443	1.41	0.58 - 3.45
Do your jaws regularly become stiff, tight or tired?	4 (14.3%)	17 (25.8%)	0.222	2.08	0.63 - 6.86
Do you have pain in or around your ears, temples or cheeks?	2 (7.1%)	9 (13.6%)	0.370	2.05	0.41 - 10.17
Do you often have headaches, neck pain or dental pain?	2 (7.1%)	19 (28.8%)	0.021	5.25	1.13 - 24.36
Have you had a recent injury in your head, neck or jaws?	0 (0.0%)	2 (3.0%)	0.352	0.69	0.60 - 0.79
Have you been aware of any recent changes in your bite?	1 (3.6%)	12 (18.2%)	0.061	6.00	0.74 - 48.59
Have you been previously treated for unexplained facial pain or a jaw joint problem?	0 (0.0%)	2 (3.0%)	0.352	0.69	0.60 - 0.79

*OR and 95% CI refer to control group as reference category; p-value denotes significance level of OR (χ^2 test)

Table 2. Prevalence of chronic pain and neuropathic pain according to sex (n = 94)

		Sex		p*
		MALE	FEMALE	
GCPS	No pain due to TMD in past 6 months	40	42	0.566
	Low disability (GI and GII)	1	0	
	High disability (GIII and GIV)	6	5	
DN4	Non-neuropathic pain	45	45	1.000
	Neuropathic pain	2	2	

*chi-squared test

Table 3. Prevalence of chronic pain and neuropathic pain according to age group (n = 94)

		Age groups			p*
		18-24 years	25-40 years	41-66 years	
GCPS	No pain due to TMD in past 6 months	33	32	17	0.526
	Low disability (GI and GII)	1	0	0	
	High disability (GIII and GIV)	6	2	3	
DN4	Non-neuropathic pain	38	33	19	0.896
	Neuropathic pain	2	1	1	

*Chi-squared test; Source: Author

There was a statistically significant difference in the nonparametric analysis of the mean age in relation to the chronic pain index (GCPS) ($p = 0.013$) with the highest degree referring to the older mean age. However, there were no statistically significant differences in the average score of the AAOP screening questionnaire in relation to the same chronic pain indices (GCPS).

DISCUSSION

Complete clefts of the lip and/or palate are immediately recognizable disruptions of normal facial structure [17]. In addition to dysfunctional facial expressions, patients with CLP may have serious functional problems with sucking, swallowing, breathing, chewing, speaking, hearing, and social integration [13]. CL/P is a highly incident malformation in newborns, occurring at a proportion of two cases per thousand live births [6]. Combined cleft lip and palate is more common than cleft palate alone. The lip and palate develop separately in the first three months of intrauterine life. This occurs due to the lack of fusion of the embryonic processes stemming from a change in the migratory velocity of the cells of the neural crest, which is responsible for the fusion of these structures. Such congenital malformations occur between the 4th and 9th week of the embryonic period due to the lack of fusion of the maxillary and medial nasal processes [18]. Functional disorders in individuals with CL/P can compromise the structures of the stomatognathic system [5], which can lead to different complaints in this population, since the different types of CL/P are closely linked to the orofacial muscles and structures adjacent to the stomatognathic system [6]. All individuals in the CL/P group (n=80) had been submitted to surgical procedures after birth to minimize the effects of abnormalities that could compromise structures of the stomatognathic system, such as occlusal deformities, hearing problems, phonological problems, difficulty breastfeeding, or other difficulties [7].

The AAOP screening questionnaire helps to identify patients who need additional clinical evaluation and is a standardized tool for research purposes. For clinical use, the patient's responses are part of the diagnosis of painful TMD. In addition, the instrument's brevity allows its routine use in clinical and research contexts, for a better assessment of patients who may have this condition [19]. A total of 80 subjects (n=80) met the inclusion criteria and answered the AAOP screening questionnaire. 53.8% (n = 43) were male and 46.3% (n = 37) were female, with an average age of 28.72 (± 11.42) years. The total of affirmative responses from the AAOP screening questionnaire and the type of CL / P and gender no significant differences. However, considering the questions in the AAOP questionnaire separately: "Are you aware of the noises in the jaw joints?" CP/L unilateral showed a more presence and relative risk compared to bilateral. This can be attributed to the neuromuscular system responsible for masticatory function has a high potential for adaptation to functional conditions. When the compensatory mechanisms of mastication and the neuromuscular system are overloaded, the result can be the occurrence of pain, joint noise or altered mandibular kinematics [2, 7, 8]. TMJ sounds are reported as "clicks" or "crepitus". It is assumed that the "click" is related to a disc displacement with reduction and the "crepitus" related to degenerative joint diseases [20]. Casanova Rosado et al. [21] argued that unilateral chronic chewing during development and growth (e.g.) can predispose an individual to internal joint degeneration and joint disorders and that unilateral can be a factor highly associated with TMD and may even be the cause of the problem. In this research the majority of individuals with CL / P were identified as unilateral when compared to bilateral, as most articles demonstrate [22, 23]. Studies comparing the type of cleft had shown that CF/L are more prevalent in the male gender and CP are more prevalent in the female gender [23, 24].

For questions the gender variable, there was a significant difference in two questions: "Does your jaw get "stuck", "locked" or "go out"?" and "Do you have pain in or around your ears, temples or cheeks?" also with male relative risk in relation to female. Population studies report a prevalence of TMD of 8% to 15% for female and 3% to 10% for male, suggesting that TMDs are significant causes of pain in the head and face, in addition to difficulty in mandibular kinematics[7, 25] but these data do not consider individuals with CL / P separately. Where a worldwide trend has been observed towards higher frequencies of cleft lip with or without cleft palate in male[6, 23, 24]. Of the total number of individuals interviewed, 75% (n = 60) answered affirmatively at least one of the questions in the orofacial pain screening questionnaire (AAOP), and the chronic pain questionnaire (GCPS) was performed for this sample. Uses of a graded classification of chronic pain have a wide range of potential uses. In epidemiological field investigators, graded classification could facilitate a more complete and reproducible differentiation of the overall severity of pain between cases. In population surveys (eg) most people report recurring pain symptoms, many report severe and persistent pain, but less are severely disabled by pain[16]. In general, a graded classification can be useful when a simple approach is needed to describe qualitative differences in the overall severity of pain. A graded classification of chronic pain should be: (1) a mutually exclusive and exhaustive set of ordered categories whose classification corresponds to qualitative differences in the severity of chronic pain; (2) based on simple measures and scoring rules to facilitate use during the clinical encounter; (3) accurate, reliable and valid, both in terms of cross-sectional association with important indicators of the severity of chronic pain and in the ability to predict patient outcomes; (4) generalizable in different anatomical sites and heterogeneous causes[26]. It is now widely accepted that ch

ronic pain is a multidimensional phenomenon. Pain intensity, pain persistence, pain-related disability and recency of onset may each be important attributes of a chronic pain condition. For selected purposes, however, a global measure of chronic pain severity is needed that summarizes different pain measures. In prior work, graded classification was offered as a possible approach to summarizing the global severity of chronic pain (GCPS)[16, 27]. Analyzing the GCPS, 13 individuals (21,5 %) of the overall sample reported not having chronic pain in the previous six month (Grade 0), whereas 33 individuals (55 %) were classified as having low disability-low intensity pain (Grade I), and as low disability-high intensity in (Grade II), 14 individuals (23,3%) were classified as high disability-moderately limiting (Grade III) and high disability-severely limiting (Grade IV)[16, 26]. When the chronic pain screening questionnaire and the GCPS were related, elderly patients are affected with a greater degree of chronic pain, these results are expected because chronic pain increases with advancing age[28]. The literature points out the importance of understanding the results of the GCPS, such as the number of days on which pain interferes with one's life and the extent of the effect on activities of daily living, work and social activities[26]. When pain symptoms persist for a long time, limitations in social or daily life should be considered to discriminate between the highest severity levels of chronic pain conditions[26, 28]. Therefore, chronic pain status classification has been used as an explanatory method for assessing the overall severity of chronic pain[26, 29, 30]. In cases of intense pain with a high degree of interference or moderate to severe disability, the interpretation is "disability due to pain"[20]. There are no previous studies on the relationship between CL/P and orofacial pain affecting the temporomandibular region. However, orofacial pain can affect any structure of the stomatognathic system and can have nociceptive characteristics that emanate from the musculoskeletal system, which is affected in a large part of patients with CL/P[31].

Regarding the type of cleft, this research found no statistically significant difference related to the score of the AAOP screening questionnaire and the degree of chronic pain (GCPS). Pain complaints were highly prevalent among the individuals with CL/P in the present study, likely due to the relationship to the adjacent morphological structures of the face[18]. A previous study reports a

relationship between CL/P and the prevalence of earaches, confirming the influence of the proximity of the structures involved; a close relationship with the muscles of mastication and adjacent structures of the stomatognathic system is found in all types of CL/P, with the possible impairment of these structures[32]. Given the proximity of the structures there may be a relationship between the affected musculoskeletal regions and CL/P, since individuals with this condition can exhibit altered muscle anatomy, as stated by Sousa and Roncalli[11]. According to Liang[12] painful TMD may be associated with mandibular asymmetry. Therefore, this disorder may be present in individuals with CL/P who have asymmetry due to the flawed formation of the structures of the stomatognathic system. The observations of other authors concerning asymmetry in patients with clefts lead to the conclusion that temporomandibular fossa lies lower at the cleft side and is steeper there[14, 33]. In the present study, the individuals with CL/P had no information on TMD. According to Slade[34] the chronicity of pain as well as the occurrence of joint noises and altered jaw movements are reasons for concern on the part of health professionals, as most individuals with orofacial pain only seek treatment after the pain becomes chronic, when there may already be an association with general health problems. Based on the present results and data reported in the literature, there is a need to repeat the investigation of individuals with CL/P using the same study objectives. Indeed, a longitudinal investigation involving adults with painful TMD found reports of persistent pain in 49% of cases during examinations performed a second time after a six-month period. Likewise, another longitudinal study found that 45% of patients with painful TMD reported pain once a week or more when reevaluated after 10 and 12 months[34]. Beluzzo[35] report that painful TMD is the most frequent orofacial pain of a non-dental origin.

Therefore, the diagnosis of this condition is of the utmost importance, as TMD is recognized as the most common chronic orofacial pain condition encountered by dentists and other health professionals. The interaction of a multidisciplinary team for the treatment of the face and adjacent structures is important to a better understanding of the problem and the establishment of specific actions in the fields of speech/hearing therapy, neurology, psychology, rheumatology, otolaryngology, dentistry and endocrinology. Patients should seek health professionals for more accurate examinations and a more precise diagnosis in each case. Indeed, the etiological differences may signify that there is no direct relationship between painful TMD and CL/P. While the former may originate from multiple factors that generally involve structural issues, the latter is classified as one of the most frequent congenital deformities[7, 18, 36]. Considering all these issues, the present findings on the prevalence of pain complaints in individuals with CL/P underscore the importance of incorporating this evaluation into the routine care for this population, as more than one patient gave at least one affirmative answer on the questionnaires used for the diagnosis of painful TMD. Further cross-sectional and longitudinal studies are needed to enable a better understanding of the possible link between cleft lip/palate and temporomandibular disorder.

CONCLUSIONS

In the present sample, the screening questionnaire that investigates painful TMD, proved to be a useful and easy tool for those with CL / P because most of the individuals in the studied group answered affirmatively to at least some questions in that questionnaire. In this condition AAOP recommends a more thorough examination by a specialist. Regarding the impact of chronic pain in relation to activities of daily (GCPS), it was shown that for most individuals with CL / P, which despite its presence, this has an impact on disability (low), regardless of its intensity (low / high) with older individuals being more limiting.

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