

Review articles

Orofacial myofunctional evaluation in cleft lip and palate: an integrative literature review

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ABSTRACT

Purpose: to identify the focus of scientific publications in the field of orofacial motricity in individuals with cleft lip and palate, as well as validated protocols used in speech and language evaluation.

Methods: a study conducted using the following databases: Medline, SciELO, Lilacs and Google Scholar, through keywords including: cleft palate + cleft lip + evaluation + speech therapy + stomatognathic system + speech + phonation + chewing + swallowing + breathing + validation studies, in Portuguese and English.

Results: a total of 572 articles that addressed the evaluation of the stomatognathic system was selected. The articles were published between 2012 and 2017, and examined for year of publication, subject area, focus, instrument used for evaluation, and type of study. Of these, 90 articles met the inclusion criteria, but only 5 used validated protocols. The greatest number of articles was published in 2014. The studies covered broad age groups, using subjective and objective methods, and instruments were often non-standardized.

Conclusion: speech was the prevalent theme, while other orofacial functions were poorly investigated, demonstrating that studies with other approaches were lacking.

Keywords: Cleft Lip; Cleft Palate; Stomatognathic System; Evaluation; Speech, Language and Hearing Sciences; Validation Studies

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INTRODUCTION

The stomatognathic system consists of oral structures and performs several essential functions that interrelate with its anatomy. Thus, any influence on this system will result in adaptation¹. Cleft lip and palate affect the stomatognathic system and, consequently, the performance of orofacial functions.

Even after surgical correction, individuals may present with orofacial myofunctional changes that require speech therapy². Thus, a detailed evaluation of the stomatognathic system^{3,4} is essential for the speech-language pathologist to diagnose and treat dysfunction. There is a need for a well-structured data collection and recording system, and for standardized evaluation protocols⁵.

Among the functions performed by the stomatognathic system, speech is altered by the presence of a cleft lip and palate. This contributes to further stigmatization of individuals with this malformation, which has been widely studied in the literature. However, since the other components of this system are also important for achieving morpho-functional balance and completion of treatment, breathing, chewing, and swallowing functions should also be addressed. The question is whether all orofacial myofunctional components are being evaluated in individuals with cleft lip and palate.

Speech therapy has emphasized the importance of evidence-based practice, and integrative review is a method that meets this objective, as it analyzes and synthesizes the results of selected studies, making them useful in both clinical practice and scientific research. This type of review has several purposes: definition of concepts, revision of theories and evidence, and analysis of methodological problems of a particular topic that allows the inclusion of experimental and non-experimental studies, in order to understand a specific subject⁶.

Therefore, this study aimed to identify the focus of scientific publications in the area of orofacial motricity in individuals with cleft lip and palate, as well as validated protocols used in speech therapy evaluation.

METHODS

This study was conducted using the following databases: Medline, SciELO, Lilacs, and Google Scholar, through keywords including: cleft palate + cleft lip + evaluation + speech therapy + stomatognathic system + speech + chewing + swallowing + breathing + validation studies, in Portuguese and English.

Articles published between 2012 and 2017 that addressed speech therapy evaluation with regard to aspects of the stomatognathic system in individuals with cleft lip and palate not associated with syndromes were selected. Articles that used any validated protocol and analysis regarding the level of scientific evidence were categorized. The titles, abstracts, and articles that met the inclusion criteria were read in full.

As a selection criterion, all literature review articles, clinical cases, monographs, theses, and books were excluded. The articles included in the study were analyzed by a single researcher and tabulated according to the following categories: year of publication, subject area, focus, instrument used for evaluation, type of study, study objective and results.

LITERATURE REVIEW

The database search identified 572 articles based on the title and abstract. Of these, 482 were excluded, 405 for not meeting the inclusion criteria and/or not involving speech therapy evaluations in orofacial motricity, and 77 due to duplication. Thus, 90 articles were included and analyzed in their entirety (Figure 1).

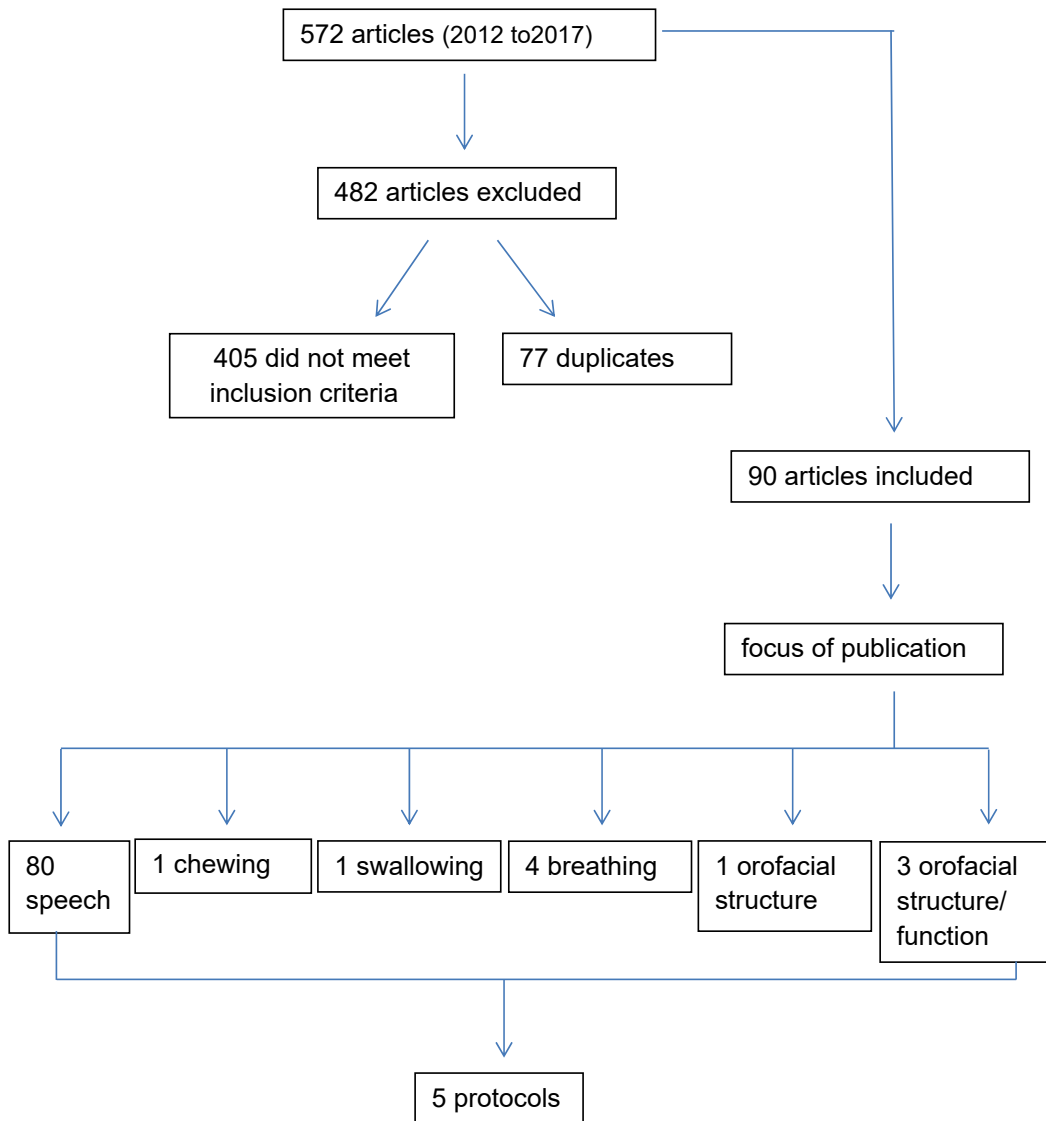


Figure 1. Steps of the literature review analysis process

The selected articles were grouped by category: year of publication, subject area, focus, instrument used for evaluation, type of study, study objective, and results. The articles were subsequently grouped according to the focus of study: 90% (80 articles) focused on the evaluation of speech, 1% (1 article) on

chewing, 1% (1 article) on swallowing, 1% (1 article) on orofacial structures, 4% (4 articles) on breathing, and 3% (3 articles) on evaluation of orofacial structures and function (Figure 2). Figure 3 shows the distribution of articles according to the subject area, and the focus of the publications by date.

Year of Publication	Subject Area	No. of Articles	Focus	Evaluation Method	Type of Study	Objectives	Results
2012 to 2017	Speech	80	- cleft lip and palate - isolated cleft palate - broad age group	- perceptual-auditory analysis - instrumental exams: • nasometry, • nasoendoscopy, • acoustic rhinometry, • rhinomanometry, • videofluoroscopy, • electromyography	• retrospective • prospective • descriptive • qualitative • quantitative	- speech evaluation • therapy, language and velopharyngeal function; - speech results after surgery	- phonetic, phonological, and velopharyngeal function-related alterations; - need for evaluation protocol and training; - effective pharyngeal flap and veloplasty in the correction of velopharyngeal dysfunction - undefined best age and surgical technique; - subsidiary instrumental methods for speech evaluation
2014	Chewing	1	- cleft lip and palate - 7 to 14 years old - 47 cases	- chewing tests - electromyography	• prospective • descriptive • qualitative	- verify concordance between 3 evaluation methods and the chewing pattern in unilateral and bilateral cleft	- unilateral fissure with greater amplitude of electromyographic signal than bilateral; - lack of agreement between methods.
2015	Swallowing	1	- cleft lip and palate - 0 to 3 years old - 7 cases	- clinical exam - videofluoroscopy	• prospective • descriptive • quantitative	- evaluate swallowing	-choking, nasal regurgitation, and failure of velopharyngeal function
2013 and 2015	Breathing	4	- cleft lip and palate - 6 to 40 years old	- clinical exam - respiratory symptom questionnaire - instrumental exams: • videonasoscopy • rhinomanometry • acoustic rhinometry	• retrospective • prospective • descriptive • qualitative • quantitative	- evaluate nasopharyngeal dimensions and respiratory symptoms after pharyngeal flap surgery	-reduction of nasopharyngeal dimension; -presence of respiratory symptoms after pharyngeal flap surgery in older individuals
2015	Orofacial Structures	1	- cleft lip - 10 to 20 years old - 70 cases	- clinical exam - nasoendoscopy	• retrospective • prospective • quantitative	- identify signs of hidden submucosal cleft in individuals with cleft lip and without clinical signs of cleft palate	-15% of cases with signs of hidden submucosal fissure
2012 and 2015	Orofacial Structure/ Function	3	- cleft lip and palate - 7 to 28 years old	- clinical exam	• retrospective • prospective • descriptive • qualitative • quantitative	- evaluate influence of the short frenulum on speech; - elaborate a myofunctional evaluation protocol applied to the cleft, and carry out content validation; - investigate speech and myofunctional alterations	- without influence on speech, but on mobility; - a protocol was developed with structural and functional aspects of the stomatognathic system, and the content was validated; - presence of compensatory, compulsory and phonological disorders; - change in tonicity, mobility (tongue, lips and cheeks), habitual posture (tongue and lips) and occlusion

Figure 2. Distribution of articles according to subject area

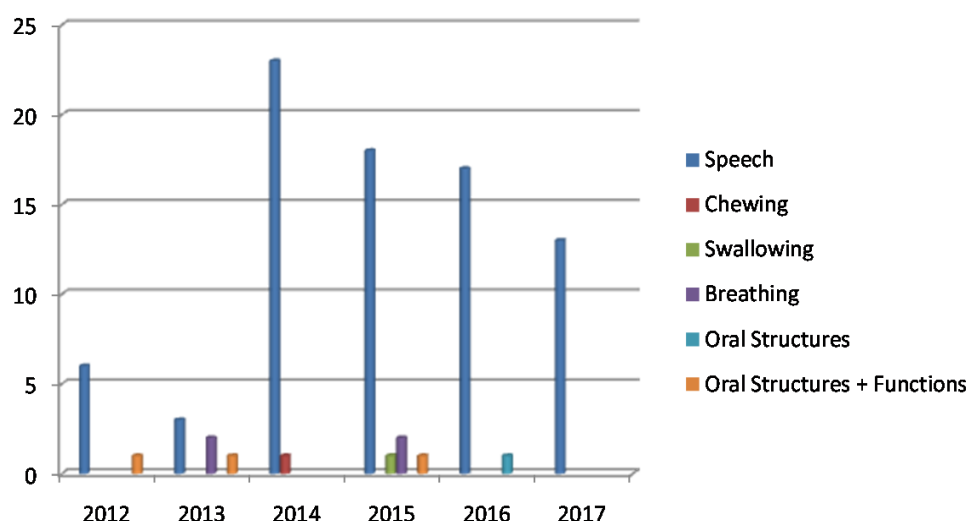


Figure 3. Distribution of research articles according to the subject area and time period

Out of 80 studies (100%) that considered speech, 26 (32%) referred to articulation, 21 (26%) to results after surgical procedures, 2 (3%) to results according to different surgical techniques, 4 (5%) to speech and language, 2 (3%) to results after speech therapy, and 25 (31%) to speech and evaluation of velopharyngeal function.

This review indicated that speech was the most discussed topic. This is understandable, since individuals with a speech impairment may be devalued by society due to their impaired ability to converse. In addition, speech challenges can influence individuals in other ways, and may be associated with emotional and/or psychological problems, such as low self-esteem, anxiety, and depression⁷, thus affecting quality of life in those with cleft lip and palate⁸.

Individuals with cleft lip and palate need repair surgery aimed at anatomical and functional correction, and studies have attempted to identify the optimal timing of surgery, as well as to define the best surgical technique^{9,10}. However, surgical failures may occur due to factors such as surgical technique and skill, cleft palate width, and/or inadequate postoperative care¹¹. Secondary surgical procedures on the palate may be necessary, mainly to provide adequate velopharyngeal function. When the velopharyngeal mechanism fails, a gap or communication between the nasal and oral cavities results in velopharyngeal dysfunction (VPD). This can be caused by a lack of tissue (velopharyngeal insufficiency) or by an alteration in the mobility of structures (velopharyngeal incompetence)¹². The effect on speech presents as compensatory articulation,

hypernasality, nasal air leakage, weak intraoral pressure, and nasal turbulence¹³.

These findings explain the prevalence of the focus on speech in the articles analyzed. Among the selected studies, 74 used perceptual-auditory evaluation in speech analysis. The literature reports that this method is the most used by speech therapists for speech analysis, and is considered the gold standard^{14,15}. However, it is a subjective method, and depends on the experience and training of the listener¹⁶. Therefore, complementary examinations are suggested to verify the reliability of the results. Complementary instrumental exams, such as nasometry, nasoendoscopy, rhinomanometry, acoustic rhinometry, videofluoroscopy, and electromyography, were identified in this literature review.

Only 5 articles used validated evaluation protocols; only 1 referred to a proposal for an orofacial myofunctional evaluation protocol, while the others used existing evaluation protocols. The other 4 studies used the *Cleft Audit Protocol for Speech-Augmented (CAPS-A)*¹⁷, which is considered a reliable, valid, and acceptable speech audit tool in individuals with cleft lip and palate. Used in the UK and Ireland, and recently by the Americleft group¹⁸, it evaluates 8 parameters of speech: intelligibility, hypernasality, hyponasality, voice, nasal air leakage, nasal turbulence, facial mimicry, and articular changes characteristic of cleft lip and palate, aiming to standardize evaluation and enable sharing of the results with other institutions¹⁹.

Using the selected articles, the characteristics of studies that presented validated evaluation protocols were analyzed (Table 1).

Table 1. List of selected articles that presented evaluation protocols in their methodology

JOURNAL	YEAR OF PUBLICATION	AUTHORS	FOCUS	EVALUATION PROTOCOL	LEVEL OF EVIDENCE
Cleft Palate– Craniofacial Journal	2014	Britton L et al.	CLP/CP (5 years old)	CAPS-A	3
CoDAS	2015	Graziani AF, Fukushiro AP, Genaro KF	75 CLP (7 and 29 years old)	CLEFT PROTOCOL	4
Cleft Palate– Craniofacial Journal	2016	Chapman KL et al.	19 CLP/CP 1 VPD (5 and 10 years old)	CAPS-A	4
Orthodontics & Craniofacial Research	2017	Sell D et al.	268 CLP (5 years old)	CAPS-A	2
Cleft Palate– Craniofacial Journal	2017	Castick S, Knight RA, Sell D	25 CLP	CAPS-A	3

Legend: CLP = Cleft Lip and Palate; CP = Cleft Palate; VPD = Velopharyngeal Dysfunction; CAPS-A = Cleft Audit Protocol for Speech-Augmented

Data on the articles presented in Table 1.

Britton, Albery, Bowden et al.²⁰:

Parameters for the analysis of speech and treatment results were established in 12 centers in Great Britain and Ireland, aiming at a national audit process to standardize the records of individuals with cleft lip and palate, and to improve treatment. This observational, cohort, prospective, qualitative study, with level of evidence 3, was split into 2 phases, and 1,110 speech samples were selected from children with cleft lip and palate born between 2001 and 2006. Samples were analyzed by speech therapists experienced in the application of the CAPS-A protocol¹⁷ to determine the optimal timing of speech evaluation and recording and to compare inter-center speech results according to established parameters: speech evaluation (2 and 5 years old), speech and surgery, and surgery and hearing. The results showed flaws in the standardization of evaluations and data recording. However, it was found that 48% of children had normal speech, 66% did not have difficulties in speech development, and 60% did not have compensatory articulation. This study enabled a revision of national speech therapy protocols, as well as modification of evaluation and treatment parameters, thus improving the ability to compare results for use in clinical practice, and to enhance the quality of life in this population.

Chapman, Baylis, Trost-Cardamone¹⁸:

The reliability of inter- and intra-rater speech results was compared in 2 studies using the CAPS-A¹⁷

protocol in British and American-Canadian English. This was a cross-sectional, prospective, qualitative, observational study, with level of evidence 4. Ten speech samples were selected from 5 and 10-year-old children with cleft lip and palate, and 9 examiners were invited to analyze them. British samples were analyzed in 3 phases: 1) before training, 2) immediately after training, and 3) one month after training; American samples were analyzed 4 to 5 months after the evaluations for subsequent comparison. Results suggested that the classification of speech results was reliable, but the study demonstrated a lack of uniformity for all analyzed parameters, which justifies prior systematic training, to ensure acceptable levels of reliability.

Graziani, Fukushiro, Genaro²¹:

This was an observational, transversal, prospective, qualitative study, with level of evidence 4. The authors developed and validated the content of a protocol entitled “Orofacial Myofunctional Evaluation for Individuals with Cleft Lip and Palate.” A total of 75 individuals of both sexes with cleft lip and palate, ranging in age from 7 to 29 years old, participated in the study and were divided into 3 life-stages, i.e., childhood, adolescence, and adult life, in order to verify the applicability of the protocol. Content validation was performed using expert opinions, as well as by the Content Validity Index, and a proposal for evaluation involving structural and functional aspects of the stomatognathic system was developed. However, the study did not use a control group to characterize

orofacial myofunctional changes or treatment results after interventions. Moreover, the protocol was not validated in its entirety.

Sell et al.²²:

Speech results and interventions performed in 5-year-old children with cleft lip and palate seen at specialized centers in the UK were evaluated. The CAPS-A¹⁷ protocol was used to evaluate the following parameters: articulation, intelligibility, velopharyngeal function, and presence of fistula. These were associated with factors such as hearing loss, speech therapy, secondary surgery, and socio-demographic and parental factors. This was an observational, cross-sectional, prospective, quantitative study, with level of evidence 2. Results demonstrated variations in speech results among the different centers in the UK. The authors observed variation in the treatment of velopharyngeal insufficiency, which indicates the importance of treatment management and early speech therapy intervention. Because this was a multicenter study, there was a great deal of variability in the interventions and a lack of standardization in evaluation, despite use of the same protocol, which probably affected the results. Some factors were not controlled, such as the choice of a single surgeon, age of reparative surgery, and surgical technique.

Castick, Knight, Sell¹⁹:

The reliability of the Visual Analogue Scale (VAS), when compared to an ordinal scale, was investigated in order to classify perceptual judgments of 6 speech parameters in individuals with cleft lip and palate: hypernasality, hyponasality, nasal leakage, nasal turbulence, intelligibility, and acceptability. This was an observational, transversal, prospective, quantitative study, with level of evidence 3. Speech therapists trained in the use of ordinal scales, CAPS-A¹⁷ protocol guides, and universal parameters²³ were invited to analyze 25 speech samples from individuals with cleft lip and palate. After hearing the samples, they classified them separately, using an ordinal scale and the VAS. Results showed that both scales are reliable instruments for all parameters evaluated. However, the use of these scales requires prior training, and the value range of the VAS may generate more subjectivity, when compared to the ordinal scale.

Although a standardized and validated protocol was generally not used for speech evaluation in the 75

remaining articles, 78% (58 articles) used perceptual-auditory evaluation, 9% (7 articles) used existing evaluation protocols, and 13% (10 articles) did not clarify the evaluation method used. It was found that 50% (40 articles) used complementary examinations for speech evaluation.

Of the 90 articles included in the study, 37% had diagnosis as their main goal, with a focus on prognosis in 63%. One study²⁴ evaluated speech intelligibility after primary palatoplasty in 12-month-old children with cleft lip and palate and verified that surgical intervention demonstrated satisfactory results for speech. Of the speech samples analyzed, 76% showed good intelligibility, 14% acceptable intelligibility, and 10% poor intelligibility. Another study²⁵ evaluated the presence of hypernasality after surgical correction of the secondary palate, and the results showed a reduction of hypernasality in 75% of cases and elimination in 32%. Of the selected studies, 90% used the judgment of experienced examiners, and 7% of lay examiners. According to the literature^{19,26}, the use of reference and training models improves the reliability of the analysis. Cross-sectional study models were prevalent (95%), and only 1 article²⁷ presented a randomized clinical study to verify the results of facial and speech development during the mixed dentition phase in individuals submitted to 2 different treatment protocols for correction of unilateral cleft lip and palate. Results showed small differences between the 2 protocols (Millard technique combined with nasal correction and Millard technique combined with nasal correction and anterior palate closure) and speech results. One of the groups showed potentially better development, while the other showed better speech results. However, neither protocol was considered superior. In 33% of the publications, the methodology and design of the study were not well defined, which highlights the need to improve the scientific method in order to enable reproducibility. The articles covered broad age groups, and complete cleft lip and palate was the most studied subject. Speech was the focus of the publications in this study period, and other orofacial functions, such as chewing, swallowing, and breathing, as well as orofacial structures such as the lips, tongue, cheeks, teeth, hard palate, soft palate, uvula, and pharyngeal walls, were poorly investigated.

Thus, studies are needed to evaluate these other aspects, as well as other orofacial functions, since changes in the morphology of the stomatognathic system present with functional manifestations. Care

standards for this population require improvement. Standardized collection and recording of evaluation results are needed, in addition to validated protocols and systematic training in their use. Therefore, new studies that consider all aspects of orofacial myofunctional evaluation with greater methodological rigor are necessary.

CONCLUSION

In the last 5 years, there has been an important increase in scientific production in the field of orofacial motricity in cleft lip and palate cases. However, speech is the prevalent theme, probably because it is the cause of greatest stigma in these individuals. On the other hand, the evaluation of other orofacial functions is still lacking despite its importance in diagnosis and in defining the overall rehabilitation process. Moreover, standardized protocols are rarely used.

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