Integral analysis of Brazilian scientific production in Orofacial Myology: state of art and future perspectives

Análise integral da produção científica brasileira em Motricidade Orofacial: estado da arte e perspectivas futuras

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ABSTRACT
A complete review of Brazilian scientific articles on orofacial myology (OM) published in the last 10 years was conducted to describe, categorize and quantitatively analyze studies in the field of OM. Data were collected from all Brazilian journals on speech-language and hearing sciences published between 2005 and 2015. All articles were reviewed and categorized according to the year of publication, type of article and subject area. Furthermore, all studies conducted on humans that reported quantitative data were analyzed regarding age group, scope, main objective and type of study. The data were discussed from the standpoint of scientific methodology applied for the improvement of methods and for evidence-based scientific development. Moreover, aspects to be considered in future studies in this field are suggested.

Keywords: Evidence-Based Clinical Practice; Subject-Based Literature Review; Myofunctional Therapy; Method

RESUMO
Esta pesquisa revisou de modo integral a produção científica brasileira em Motricidade Orofacial nos últimos 10 anos e teve como objetivo realizar uma descrição e categorização geral das publicações em motricidade orofacial e analisar os estudos quantitativos na área. Os dados foram coletados em todas as publicações em motricidade orofacial publicadas nos últimos dez anos, após uma análise integral de todos os periódicos brasileiros em Fonoaudiologia. Os artigos foram categorizados quanto ao ano, tipo de artigo e área temática. Além disso, foram analisados sobre a faixa etária, abrangência, essência do objetivo e tipo de estudo todos os artigos realizados com seres humanos e com dados quantitativos. Os dados foram discutidos sob a ótica da metodologia científica aplicada ao aperfeiçoamento do método e desenvolvimento da ciência baseado em evidências. Além disto, os dados são analisados com sugestões de aspectos potenciais a serem considerados para futuras pesquisas na área.

Descritores: Prática Clínica Baseada em Evidências; Literatura de Revisão como Assunto; Terapia Miofuncional; Metodologia
INTRODUCTION

The last few decades have been characterized by technological breakthroughs and the concomitant progress of scientific research in the biosciences. Speech-language and hearing sciences were recognized as their own field, spurring growth in scientific studies on human communication\(^1\). Additionally, health practices became evidence based, giving rise to research on speech therapy that specifically addressed prevention and rehabilitation practices\(^1\).

Orofacial myology (OM) is the area of the speech-language and hearing sciences responsible for the study, diagnosis and rehabilitation of the structural and myofunctional aspects of the orofacial and cervical areas related to the stomatognathic functions of speaking, sucking, breathing, chewing and swallowing\(^2,3\). The first Brazilian study on OM was published by the speech therapist Beatriz Alves de Edmir Padovan in 1976. It addressed the diagnosis and treatment of atypical swallowing and was presented at conferences in the area of orthodontics in the state of São Paulo\(^4\).

A previous literature review evidenced a growing number of studies on OM published in scientific journals from 1970 to 2000\(^4\). In the last ten years, the area has exhibited significant developments, particularly during the movement to separate dysphagia as a specialty area of the speech-language and hearing sciences in Brazil. Before this movement, dysfunctions in all aspects of the physiological process of swallowing were a part of OM. This area was included in a review between 2000 and 2005, which observed that OM had less than 10% of Brazilian production with impact in communication disorders\(^5\). Furthermore, the authors reinforce the need to promote more equitable distribution of topics, as well as the expansion of research by age groups and types of intervention\(^5\).

During that time, several OM practices were established and various precepts were strengthened, while others were quickly incorporated into clinical practice in the speech-language and hearing sciences. Furthermore, the close relationship between the speech-language and hearing sciences and dentistry became more evident\(^6,7\), most likely in response to the topics addressed in the clinical practice of OM and dentists’ need for complementary alternatives to treat several conditions.

The last reference about scientific production on speech-language and hearing sciences impact factor describes low average citation per article, suggesting the lack of synchrony between the publications produced in this country\(^8\). The development of research indicates the growth and expansion of the knowledge produced by any field of study. Furthermore, the basis of clinical and social applications also stems from scientific production. Thus, the objective of the present study was to determine the state of the art of Brazilian scientific production in OM and to discuss suggestions based on the observed parameters.

METHODS

An extensive systematic literature search was conducted by four independent researchers trained in the relevant study methods and in scientific research. The search included all active and discontinued Brazilian journals in the field of speech-language and hearing sciences. All volumes of journals published online were selected, and each researcher individually identified studies whose main study object was framed in the area of OM.

Phase 1 - General categorization of publications on OM

In this phase, the abstracts of all articles contained in all volumes of journals published from January 2005 to March 2015 were analyzed. There were no language restrictions. Figure 1 summarizes the number of journals and volumes analyzed.
The following eligibility criteria were adopted for publications to be included in the general categorization phase:

- Being published in an active or discontinued scientific journal in the field of speech-language and hearing registered with a code in the International Standard Serial Number (ISSN) Network;
- Having a main study object framed in the area of OM.
- No publication whose study object included swallowing changes was considered for analysis in this review, in compliance with the principles of Resolution No. 382 of the Federal Council on Speech-Language and Hearing Sciences (Conselho Federal de Fonoaudiologia), which recognizes dysphagia as a specialty area. However, publications addressing aspects involved in the normal physiology of swallowing were included in the review because swallowing is a component of stomatognathic function (main study object in the area of OM).

The following variables were collected during the analysis of the published studies for the general categorization of scientific production: (a) journal name, (b) year of publication, (c) type of article and (d) main topic. All disagreements over studies were resolved by consensus among the four researchers.

Due to the discrepancies observed among publications indicating the subject areas of OM (clinical or research scope), 9 areas of scientific research on OM were compiled and described (Figure 2).

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**Figure 1.** Brazilian scientific journals in the field of speech therapy published through March 2015.
type of study was assessed using a research design algorithm described in Kennely et al.\textsuperscript{11} (Figure 3).

Because of the numerous discrepancies observed between the description of the type of study reported in the article and its actual classification, the type of study was determined by the researchers themselves during the analysis of full texts based on the method described by the authors of the articles.

Descriptive statistical methods were used for data analysis using the Statistical Package for Social Sciences (SPSS) version 20.0 for Windows (IBM Corp., Chicago, USA).
area of OM met the inclusion criteria. The distribution of publications between January 2005 and March 2015 is shown in Figure 5.

Figure 3. Assessment of adequate study designs based on the objective’s essence according to Kennely et al.11.

RESULTS

Figure 4 outlines the research phases and application of the study selection method. A total of 366 scientific articles published in Brazilian journals in the area of OM met the inclusion criteria. The distribution of publications between January 2005 and March 2015 is shown in Figure 5.

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* Qualitative studies were not analyzed in detail in the second phase of the study.

Figure 4. Application of criteria and article selection in the research phases.
Analysis of scientific production in myology

Figure 6 illustrates a numerical curve of OM publications in the past decade, indicating an increase in publications in the field from 2010 to 2015.

The analysis of the types of scientific articles indicated that publications on OM corresponded to 14.33% of the scientific production of the analyzed scientific journals in the field of speech-language and hearing sciences. A predominance of original articles (73.53%) was observed. Figure 7 shows the distribution of article types arranged based on a scientific evidence pyramid.

Based on the subject areas, approximately 40% of the publications on OM from the last decade addressed aspects related to the study and diagnosis of normal morphological and functional patterns of stomatognathic functions, which were included in the field of craniofacial morphophysiology. The frequency distributions of the other areas established in the present study are shown in Figure 8.

According to the age group described in each study, the most researched groups were children and adults (40.7% and 33.9%, respectively). Newborns, adolescents and the elderly accounted for 17.9%, 15.6% and 6.8% of the age groups researched in OM. Six percent of the studies did not describe their methods and results, precluding the identification of an age group.

The analysis of original research studies on OM also showed that 79.2% (n=205) of the studies had a clinical or local scope, and only 16.6% (n=43) were population based. The scope classification could not be determined in 4.2% of the articles.

Based on the main objectives of the research articles, studies assessing the prevalence of diseases or injuries were the most common (65.3%), followed by studies conducted in the field of diagnosis (15.5%) and prognosis (4.8%). Furthermore, 88% of the study designs (types of study) adopted in the articles were adequate based on their main objective. Cross-sectional studies accounted for 86.1% of the publications analyzed. Figures 9 and 10 show the distribution of articles on OM according to their main objective and type of evidence, respectively.
Figure 7. Percentage of the types of articles on orofacial myology organized based on the scientific evidence pyramid, n=374; %(n).

Figure 8. Percentage of original research articles by the subject area of orofacial myology, n=374; %(n).
DISCUSSION

Current research in biomedical sciences increasingly requires the production of evidence-based knowledge. We refer to the term evidence as the knowledge produced by controlling factors that may affect how the phenomenon studied is analyzed. Accordingly, scientific methods able to meet clinical demands must be adopted and should, whenever possible, incorporate methodological strategies to ensure population representativeness, providing scientific evidence in a wide range of clinical outcomes and enabling the proposed hypotheses to be adequately tested.

General analysis of the articles on orofacial myology

It was clear that OM subjects garnered interest among the Brazilian speech-language and hearing sciences community, as 14.33% of the publications addressed this field. Furthermore, an increase in the number of publications was evident in the period evaluated. Thus, the greater the production of knowledge, the greater the trend towards the improvement of research techniques and methods, which allows for the critical analysis of the scientific literature.
The results indicate that the journal Revista CEFAC is the most significant Brazilian research journal in the area of OM, corroborating the findings of Alves in the decades between 1970 and 2000. The curve of the annual number of publications on OM (Figure 6) illustrates a large drop in the number of publications in 2013, suggesting a possible inconsistency in the frequency of manuscripts submitted for peer review.

Nearly 25% of the publications on OM are in the lower strata of the evidence-based pyramid, and nearly 15% of these articles have no scientific evidence (Figure 7). This finding highlights the need for methodological implementation in research, as ¼ of the scientific publications in the last decade are based on models without scientific evidence. Moreover, considering that one of the roles of health research is to implement and update the procedures used by clinicians in general, exposing healthcare professionals to this type of scientific production without methodological rigor may compromise the theory in the area and the strategies chosen by professionals in their practice.

This review identified that most current research topics focus on the identification of normal aspects of craniofacial organs (craniofacial morphophysiology). Among the more specific topics, studies on neonatology published in recent years are well represented.

When analyzing the practice areas determined by the Brazilian Speech Therapy Boards, we observed low scientific production in the areas of facial esthetics and gerontology, despite the theoretical and technological advances in recent years. Both areas have important social applications in OM and are potential catalysts of the development of this science, especially gerontology, in light of reports of increased life expectancy and reduced population mortality in the age group older than 60 years.

The low number of studies in gerontology was confirmed by the observation that the age group least researched in the last decade in OM was the elderly, with less than 7% of the total number of publications.

Analysis of quantitative OM studies: Main objectives

Approximately 65% of the quantitative studies on OM are focused on determining prevalence, followed by diagnosis (15.5%) and treatment (6.8%). Furthermore, the data show a low number of OM studies on the negative impacts of disorders and dysfunctions (impairment/loss), the natural history of OM changes (etiology), the occurrence of new cases of diseases (incidence) and estimates of the clinical progression of OM changes (prognosis). These data may be partly explained by the low percentage of population studies published in the decade analyzed.

The need for scientific production on the etiological determination of changes studied in OM was previously highlighted in an earlier review of articles from the 1970s to 2000. However, the same weaknesses persisted during the last decade. Other aspects of basic research studies requiring further detailing according to this review included the natural history of disorders and dysfunctions and prognosis and diagnosis in OM. An example of the current state of research is the need for the objective quantification of the long-term effects of different anatomical patterns (congenital or acquired) on the functional aspects of the stomatognathic system. The present findings show that only 5% of the publications in OM in the last decade with scientific evidence reported a method allowing this type of conclusion.

The imbalance observed in the coverage of population studies reveals a weakness in OM research even today, which limits the reliability of the data and precludes extending the findings to other populations. Given the current process of stabilization of the area, following the separation of dysphagia, the description of epidemiological parameters is critical for the knowledge of the distribution and definition of aspects involving the normal and disease patterns and their biological, social, age-related and regional variations, among others. The need for OM research to expand its clinical scope and academic space towards the general population is evident, as reported by Alves.

Epidemiological research aims to describe the distribution of health conditions, the occurrence of diseases, the prediction of their frequency and the control and prevention of the onset of new cases. Population surveys must be performed to quantify the impact of the disease or dysfunction in question, thus contributing to evidence-based practice and the prevention and reduction of the impact of diseases. Furthermore, such studies have a key role in supporting public health policies on speech therapy and are the basis for raising public funds for the area.

Because healthcare intervention at any level has the power to change the epidemiological profile of social groups and affect health or disease determinants, epidemiological research much be encouraged to increase the population’s access to speech-language and hearing sciences, particularly the specialty of OM. The use of epidemiological data for prioritization, resource allocation and programmatic guidance is one of the principles guiding the Brazilian Unified Health
System (Sistema Único de Saúde, SUS)\textsuperscript{19}. Furthermore, the epidemiological approach is the primary strategy for the implementation of the national health promotion policy\textsuperscript{20}. Accordingly, the results of this review underscore the argument of Goulart \& Chiari\textsuperscript{21} on the need to implement population surveys able to provide more detailed information on the characteristics of individuals who are susceptible to communication disorders, particularly those related to the functions of the stomatognathic system (SS).

Due to the characteristics of a population study, including human and financial resources, several Brazilian and international research centers contributed to expanding these resources by developing multidisciplinary and multi-centric research strategies with well-defined and validated criteria. Despite the national distribution of OM research centers, the appropriate selection of research hypotheses and the use of valid criteria combined with joint research teams may be a promising strategy to advance the field of OM in Brazil.

Analysis of quantitative OM studies: Types of studies

Considering the scientific evidence of research findings, the chronological relationship between data measurement and the occurrence of the phenomena studied becomes relevant. The time pattern of a study is directly related to the quality of the data collected\textsuperscript{17}, which is best when collected longitudinally. The results of this review show that over 85\% of the quantitative studies published in the last decade in the area of OM were cross-sectional and were mostly focused on prevalence designs, instead of clinical trials and cohort or case-control studies. This finding highlights the need to deepen and develop the methodologies applicable to longitudinal studies in OM research. Thus, the previously examined conceptual hypotheses could be analyzed again with an ongoing perspective to provide a greater power of evidence.

Given the high frequency of cross-sectional studies, the data show the abundance of methodological confounding variables, termed biases, in the literature. The number of biases is directly proportional to the study design, and the closer to the base of the evidence-based pyramid the study is, the larger the bias will be. Accordingly, inferences made by a single clinician and published as a case series, for example, are at the lowest level of the evidence-based pyramid and are therefore more susceptible to biases. The next level of evidence includes observational studies that measure some exposure and outcome, such as cross-sectional, case-control and cohort studies, in ascending order\textsuperscript{23,12}.

Considering the intervention variables in the outcome of a study with humans, clinical trials (with or without randomization) are the next level of evidence, and the strongest level of evidence is provided by systematic reviews (with or without meta-analysis), which combine the results of clinical trials with different publications and compile their findings.

Many research questions were examined in recent years, albeit mostly using designs with a low level of scientific evidence. Therefore, the area of OM has a wide range of conclusions that may be further questioned by researchers to confirm or modify them using methods and experimental designs with greater power of evidence.

During the stipulation of the study hypotheses, the selection of an appropriate study design must be considered a critical point of the study. Furthermore, the researcher should be aware of the type of conclusions each design supports to avoid over- or underestimating the data or inappropriately judging the relationship between the variables studied. Using data from cross-sectional studies to explain causal relationships between events is a commonly observed practice\textsuperscript{17}. Differently from general conceptual hypotheses, to determine a cause-effect relationship of a condition, it is necessary that the data be reproduced in several studies and different populations and not used to merely identify statistical significance.

Conceptual hypotheses based on clinical practice and observation are confirmed (or refuted) upon systematic observation of the researched phenomena. The study design and execution must be well planned for the study conclusions to have merit. In the last decade, nearly 12\% of the quantitative OM studies had inadequate experimental designs. Clarity and objectivity in the selection of hypotheses to be tested in a study are essential to identify the appropriate design that will produce reliable results that express the reality observed.

Future perspectives

Historically, speech therapy in OM was empirically and subjectively performed in the evaluation and treatment of orofacial structures\textsuperscript{22}. Furthermore, the difficulty in establishing parameters facilitating diagnosis and in explaining the prognosis of patients is evident\textsuperscript{23}. The analysis of the quality of studies in OM shows weaknesses similar to those mentioned and may represent the greatest challenges and priorities of OM research today: the classification and definition of
OM dysfunctions and disorders based on longitudinal studies, the determination of valid and easily reproducible diagnostic instruments and the assessment of the therapeutic efficacy of intervention protocols.

The field of speech-language and hearing sciences must organize the data on OM and design appropriate criteria for therapeutic methods, as stated by El-Dib & Atallah24. The subsequent step in defining diagnostic criteria is the comparison of intervention protocols, ideally including randomized clinical trials. These clinical trials will confirm the effectiveness of therapy while controlling biases.

Evidence will provide researchers the tools and human and financial resources necessary to expand this specialty in public health policies. However, evidence should be gathered through clinical trials as required by the Brazilian legislation19. The expansion of public policies and job opportunities in the field of speech-language and hearing sciences is directly related to the development of evidence-based technologies and their use by health professionals1. The development of the field and the dissemination of knowledge during professional training are the roles of research centers, non-profit organizations in the speech-language and hearing sciences and universities (particularly public universities).

Improving the scientific method and standardizing the assessment and diagnostic criteria are key points to be prioritized with urgency. Complementary assessments and low- or high-complexity examinations have the ability to contribute to both diagnosis and treatment planning5. Nevertheless, the current use of valid, objective and easily reproducible instruments (for screening or diagnosis) is still scarce in the area of OM. Consistent and reproducible evaluation methods should be used in research on human communication and its disorders29 because their absence decreases the faithful reproducibility of diagnostic and therapeutic practices, directly affecting study conclusions and evidence produced in the area. Accordingly, a system for the evaluation and classification of valid alterations that can, at least in theory, be reproduced in other populations by any expert in the field is crucial.

More broadly, the development and validation of assessment instruments must be achieved with discretion. Such instruments must be able to determine the number of true positives among those testing positive. Currently, the methods and instruments with wide applicability, such as the evaluation of facial muscle function, are not able to predict these parameters. Even a validated instrument with high sensitivity and specificity may generate erroneous data by failing to correctly identify the individuals evaluated. This dilemma is only overcome when the prevalence of disease in the population is known so that the quality of the instrument may be assessed by calculating the "positive predictive value"25,26. The need for this type of approach in research in the speech-language and hearing sciences was previously mentioned by Goulart & Chiai1, and the current findings express the lack of these resources in OM.

Conducting research on OM using patients should be the basic research standard to clarify the clinical characteristics related to the etiology and prognosis of SS dysfunctions or disorders and to report scientific evidence for clinical interventions. The intellectual investment in research planning with a higher quality of evidence becomes essential, starting with updating the theoretical foundations of the instruments and their parameters, subjecting protocols to comprehensive pilot studies and standardizations and forecasting the effects, conditions and duration of studies.

Planning and performing case-control studies will ensure results that may coherently introduce discussions on health impairments caused by the alterations of the SS, confirm diagnoses and describe the origin and incidence of various changes. Cohort studies, when properly conducted, will produce impactful results for the scientific community about the natural history of changes in the SS, for which evidence in the literature on OM is still scarce. Furthermore, cohort studies and clinical trials in OM will allow for the temporal determination of therapy progress and confirmation of the efficacy of intervention protocols with pre-established timelines.

Such data will enable the scientific community in the coming decades to establish the duration of the onset period of some disorders and dysfunctions and the time required for an intervention to have positive effects on a patient; it may also allow for the estimation of the time to hospital discharge for some conditions.

The combination of theory- and evidence-based practices will enable OM specialists to become increasingly confident and independent, to develop a more systematic clinical rationale, to better choose treatment strategies and to achieve effective results. These values will certainly affect how the specialty is perceived by society.
CONCLUSIONS

The present systematic review showed an increase in the number of publications in the area of orofacial myology in the last decade. Furthermore, regarding methodological aspects, most quantitative data in the area were collected in cross-sectional studies.

The scarcity of scientific production in OM encompassing the etiological determination of alterations, the natural history of disorders and dysfunctions and prognosis and diagnosis was clear. Furthermore, these findings indicate the need for well-developed methods that may be used in longitudinal studies to strengthen the available knowledge and the status of the field of speech-language and hearing sciences in the health sciences community through the multidisciplinary approach of evidence-based practice.

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