



Intellectual disability: scientific literature about orofacial myofunctional characteristics

Deficiência intelectual: produção científica acerca das características miofuncionais orofaciais

Discapacidad intelectual: producción científica sobre las características miofuncionales y orofaciales

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Abstract

Introduction: Intellectual disability is associated with multiple etiologic factors, with reports of structural and functional alterations of the stomatognathic system. **Objective:** To review the literature regarding the scientific production and the characteristics of orofacial myofunctional disorders of people with intellectual disabilities. **Method:** National and international journals were analyzed in Lilacs',

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Medline's, Scielo's and Google Scholar's databases between 2000 and 2012, by searching for terms like oral motricity, mental disabilities, tongue, lip, swallowing and chewing, being excluded those who did not mention if the individuals in the sample had or not some intellectual disabilities. **Results:** On the characteristics of the scientific production, there was a prevalence of clinical studies with description of syndromic cases, the age of the individuals studied was between six months and 67 years (greater focus on childhood) and prevalence of male gender. The most described type of intervention was the evaluation / diagnosis, with higher production in 2010. Despite the heterogeneity in the studies, the changes cited were: in swallowing (in 80% of selected texts), in orofacial muscle tone (64%), structures of the stomatognathic system and speech (60% each), in posture and mobility of the articulators (48% each) in chewing (44%) and in breathing (36%). **Conclusion:** The Brazilian production has investigated, with greater commitment in the last decade, the association between orofacial myofunctional disorders and intellectual disabilities, especially in clinical syndromic condition and in all researches were cited structural or functional abnormalities of the stomatognathic system, suggesting that studies in this area are essential for a better understanding of the issues involving the neurology, anatomic and physiopathologic aspects in this disability.

Keywords: mastication, deglutition, respiration, intellectual disability, stomatognathic system.

Resumo

Introdução: a deficiência intelectual está associada a vários fatores etiológicos, com relatos de alterações estruturais e funcionais do sistema estomatognático. **Objetivo:** analisar a literatura quanto à produção científica e às características dos distúrbios miofuncionais orofaciais de pessoas com deficiência intelectual. **Método:** foram analisados periódicos nacionais e internacionais nos bancos de dados Lilacs, MedLine, Scielo e Google Acadêmico entre 2000 e 2012, por meio dos termos motricidade oral, deficiência mental, língua, lábio, deglutição e mastigação, sendo excluídos aqueles que não mencionavam se os indivíduos da amostra apresentavam ou não deficiência intelectual. **Resultados:** Quanto às características da produção científica houve prevalência de estudos clínicos com descrição de casos síndromicos, a faixa etária dos estudos compreendeu sujeitos entre seis meses e 67 anos (maior enfoque na infância) e com prevalência do gênero masculino. O tipo de intervenção mais descrito foi o de avaliação/diagnóstico, com maior produção em 2010. Apesar da heterogeneidade dos estudos, as alterações citadas foram: na deglutição (em 80% dos textos selecionados), na tonicidade orofacial (64%), nas estruturas do sistema estomatognático e fala (60% cada), na postura e na mobilidade dos órgãos fonoarticulatórios (48% cada), na mastigação (44%) e na respiração (36%). **Conclusão:** A produção brasileira tem investigado, com maior afinco na última década, a associação entre os distúrbios miofuncionais orofaciais e a deficiência intelectual, principalmente em quadros clínicos síndromicos e em todas as pesquisas foram citadas alterações estruturais ou funcionais do sistema estomatognático, sugerindo que estudos nessa área sejam fundamentais para melhor compreensão dos aspectos neuroanatomofisiopatológicos envolvidos nessa deficiência.

Palavras-chave: mastigação; deglutição; respiração; deficiência intelectual; sistema estomatognático.

Resumen

Introducción: La discapacidad intelectual se asocia a múltiples factores etiológicos, con relatos de alteraciones del sistema estomatognático. **Objetivo:** analizar la literatura en relación a la producción científica y las características de los trastornos miofuncionales orofaciales de las personas con

discapacidad intelectual. Método: se analizaron periódicos en las bases de datos Lilacs, Medline, Scielo y Google Académico (2000 y 2012), a través de los términos motricidad oral, discapacidad mental, lengua, labio, deglución, masticación, siendo excluidos aquellos que no mencionaban si los individuos del muestreo presentaban o no discapacidad mental. Resultados: En cuanto a las características de la producción científica, hubo prevalencia de estudios clínicos con descripción de los casos de síndromes, la edad de los sujetos de estudio estuvieron comprendidas entre los seis meses y los 67 años de edad (mayor enfoque en la infancia) y la prevalencia del sexo masculino. El tipo de intervención más descripto fue la evaluación/diagnóstico, con mayor producción en 2010. A pesar de la heterogeneidad de los estudios, los cambios citados fueron: en la deglución (en 80% de los textos), en el tono orofacial (64%), en las estructuras del sistema estomatognático y del habla (60 % cada), en la postura y en la movilidad de los órganos fonoarticulatorios (48%), en la masticación (44%) y en la respiración (36%). Conclusión: En la producción brasileña se está investigando, con más dedicación en la última década, la asociación entre los trastornos miofuncionales orofaciales y la discapacidad intelectual, especialmente en los cuadros clínicos de síndromes. En todas las investigaciones fueron citadas alteraciones estructurales o funcionales del sistema estomatognático, lo que sugiere que estudios en este área sean esenciales para una mejor comprensión de los aspectos neuroanatomofisiopatológicos involucrados en esa discapacidad.

Palabras clave: masticación, deglución, respiración, discapacidad intelectual, sistema estomatognático

Introduction

Intellectual disability (ID) entails restrictions in different aspects of people's life that have it, as in learning, in interpersonal relationships, in occupational and in social activities, depending on the dimensions of the disability, the severity and the environment in which the subject is inserted¹.

It follows different syndromes that can affect the orofacial myofunctional functions in syndromes like Cornelia Lange², Fragile X³, Down (DS)⁴⁻⁹, Silver-Russell¹⁰, Goldenhar¹¹, Angelman¹²⁻¹³, Worster-Drought¹⁴, Sturge-Werber¹⁵, in congenital myogenic¹⁶ among others.

Other conditions may also have association with ID as, for example, in not progressive encephalopathies (NPE)¹⁷⁻¹⁹, in Lesch-Nyhan²⁰ disease, in neurofibromatosis type 1²¹⁻²², in mucopolisacaridoses²³, and other diseases with unknown causes²⁴⁻²⁶.

Due to the diversity of possibilities of presence of ID alone or associated with other changes, the central questions of this study were related to trends of research in oral motricity (OM) and the possibility of the existence (or not) of orofacial myofunctional disorders in people with this disability. Therefore, this study aimed to analyze the scientific literature about the production and about the characteristics of orofacial myofunctional disorders of people with intellectual disabilities,

in order to justify (or not) speech therapy in this area, as well as to guide the professional practice in OM on scientific evidences.

Method

A qualitative systematic review research in which were analyzed journals found in LILACS', MEDLINE's and SCIELO's databases between 2000 and 2012, totaling 12 years of scientific publication.

The initial procedure of data collection occurred from the selection of articles with the term "oral motricity". The selection of the articles was conducted in Portuguese (Brazil and Portugal), Spanish and English. 55 results were obtained, and were included those that treated orofacial myofunctional disorders in individuals with ID and excluded those who did not cite or describe them in method's chapter, if the individuals in the sample presented or not the respective disability.

Thus, three articles were obtained. Due to the reduced number of items, keeping inclusion and exclusion criteria mentioned above, the search extended in two ways: 1) The research was taken up in the LILACS', MEDLINE's and SCIELO's databases, adding the keywords: tongue, lip, chewing and swallowing, linking them to intellectual disability and 2) Inquiry in Google Scholar's database (up to page 10 of searching) with the associated

terms: mental disability and oral motricity. Since the survey was conducted in two databases, it was necessary to compare results, eliminating from the research the repetitive and articles that were already selected.

Thus, 25 results were obtained by proceeding with the entirety articles reading, and the recognition of each article's features. For this goal, two researchers (P1 and P2) performed the search and critical reading of the selected articles independently, considering some analysis parameters described below.

For the analysis of the articles it was built a record containing information that would be marked with multiple choices, being selected the following analysis variables of the characteristics of the studies reviewed:

- *Type of production*: we considered the theoretical (systematic literature reviews and meta-analysis with inclusion of Cohort studies, control groups, integrative reviews and criticism), clinical studies (randomized, series of cases and case reports), cross-sectional surveys (with or without case-control and semi-experimental) to make it possible to analyze the scientific evidence obtained from the publications.

- *The individuals' age and gender*: with blanks to fill with the individuals' age and gender, except the theoretical papers (if they existed);

- *Type of intervention*: procedures of triage were selected, diagnostic / evaluation, therapeutic procedures and it was noted if the intervention occurred or not in an interdisciplinary way;

- *Changes description in OM*: it was selected alterations related to structural and postural characteristics, reflexes, sensibility, muscle tone, mobility, speech, breathing, chewing, swallowing and dentition were selected.

- *Level of intellectual disability*: it was classified as mild, moderate or severe.

For the analysis of the results it was considered in the articles: the number of articles produced by

the journals consulted and the types of production; regarding research: findings of speech therapy interventions related to orofacial myofunctional diagnosis; among the subjects investigated were: age, gender and level of intellectual disability.

P1 and P2 met and discussed the congruence or not of the inclusion and an analysis parameters interpretation, and when disagreed, a third researcher (P3) pondered the doubt, coming to a consensus.

The obtained data was synthesized and initially distributed according to the characteristics of the articles and, therefore, they were tabbed in Excel spreadsheet software (Microsoft® Office package) to a descriptive data analysis, performing simple arithmetic sum and percentile analysis. No statistical method was used, bearing in mind that it was intended to combine the results of two or more studies. After the articles first phase characterization of the obtained results, it moved to the second phase, an orofacial myofunctional disorders general analysis reported by articles and the third stage consisted in performing syntheses presented by the condition.

Results

Regarding the type of bibliographic production on the investigated subject, from the 25 articles analyzed, ten (40%) were cross-observacionais^{4,5,9,14,17,20,22-24,26}, nine (36%) clinical case reports^{2-3,10-12,15-16,18,21} and six^{6-8,13,19,25} (24%) literature review. The highest concentration of publications occurred in 2010, with five studies published this year. It is noteworthy that from the 25 articles, the biggest part of them^{2-4,7-13,15,17-19,21-26} (20-80%) belonged to Brazilian researchers. Regarding scientific evidence of the analyzed articles, the results can be seen in Table 1.

Hierarchical Order	Description	
1 - Theoretical	With meta-analysis of Cohort studies and clinical trials with Control Group	No publication found with such characteristics.
	Integrative systematic reviews	Two publications ^{8,13} , one on evaluation and therapy in Down Syndrome (DS) ⁸ and another one on the evaluation of Angelman ¹³ Syndrome.
	Critical or narratives reviews	Four publications ^{6,7,19,25} , one from Portugal ⁶ which described the odonto-maxillofacial alterations, oral diseases and deleterious oral habits in persons with Down syndrome and a monograph describing the dental and orthodontic aspects in DS ⁷ . The NPE was the subject of a literature review of dentistry being contemplated orofacial myofunctional disorders ¹⁹ . Without specifying the source of ID, Brazilian researchers conducted ²⁵ the theoretical review of these subjects' oral health status.
2 - Clinical Trials	Randomized and with the control group	No publication found with such characteristics
	Longitudinal studies	Angelman syndrome description from a clinical case ¹² and a 3: 8 years old ¹⁸ boy's NPE.
3 - Transversal Studies	Homogeneous groups (same condition)	The DS was quoted in Brazilian article describing the results of orofacial myofunctional evaluation of four children aged up to four years old ⁹ . London researchers evaluated 42 individuals with a diagnosis of Worster-Drought syndrome ¹⁴ . Brazilian researchers ¹⁷ evaluated 32 individuals with NPE, aged between 1: 6 and 8: 3 in order to verify swallowing. A multicenter study ²⁰ described the characteristics of 44 subjects with Lesch-Nyan disease (aged from 2 to 38 years old) treated for 3 years. From retrospective study ²² (analysis of 183 records), it was the description of the general characteristics of speech therapy and subjects with neurofibromatosis type 1. A study from Rio Grande do Sul ²³ characterized the system and stomatognathic functions in 78 patients (aged between five and 21 years old) with mucopolysaccharidosis.
	With Control group	Only one publication (no description of the method) ²² .
	With statistical analysis and description of the confidence interval	Already described Publications ^{14,22} and in a dental-surgeons' study from Rio Grande do Sul ²⁴ in which 138 documentations of individuals with disability were analyzed (various ages, from babies to the elderly).

Table 1 - Scientific evidence source articles between 2000 and 2012 about intellectual disability and oral motricity.

The study age range included individuals between six months old and 67 years old (the majority of studies involving individuals with disabilities investigated infants, preschoolers and children, i.e., the age group of zero to 12 years old was prioritized - with 16 publications - 64%). About the gender, in the articles which were presented as clinical reports and cross-sectional studies, the incidence was higher in males (395 cases - 59.94%) than females (264 to 40.06%).

Regarding the type of intervention, most studies (21 - 84%) involved evaluation / diagnosis (medical or phonoaudiological)^{2-11,13-17,20-24,26} and treatment was described in seven^{7-8,12,18-19,24-25} studies (28% - some were together with evaluation^{7-8,24}). Furthermore, in 16% of the studies^{2,4,10,19} the description was interdisciplinary.

The researches which involved OM in people with ID were characterized, with higher prevalence, for syndromic cases (14 - 56%), and among the

syndromes presented, the most cited (six publications) was Down syndrome. Non-syndromic patients were reported in eight articles (32%) and three (12%) articles did not specify the diagnosis.

About the general analysis of orofacial myofunctional disorders, it can be seen that the structural alterations of the stomatognathic system were cited in 15 publications (60% of authors)^{2,3,6-13,15-16,21,22,25}, with the following changes, in descending order of appearance: change in size, shape or volume of the hard palate was cited publications in eight (32%)^{2,3,6,9,11,15,16,21}; head^{6,7,10,12,13,16,22}, face^{3,6,9,10,11,15,16}, mouth and lips^{2,6,9,11,13,15,21} (7 items each, corresponding to 28%); maxillo-mandibular discrepancies in six articles (24%)^{2,6,8,15,16,21}; tongue (Articles 4, 16%)^{6,10,13,15}; corporal^{10,11,22} imbalances and alterations in the size or shape of the nose^{2,6,15} in three publications (12%); changes in the implantation or ear format in two articles (8%). In addition, changes in posture (of speech and body organs) were cited by 12 articles (48% of publications)^{3,6,8-13,15,18,19,23}, since the altered position of the tongue (10 - 40%: anteriorized^{3,8,12,13,15}; interposed^{6,18,23} and floor of mouth^{3,9,10}) and her lips parted in rest^{3,6,9,10,11,12,15,18,19} (nine - 36%) were the most common manifestations. The mandible deviation, quoted in two (8%) articles^{15,18} (without specifying whether such deviation occurred at rest, during mobility or execution of oral functions) and the forward position of the shoulders and the head (an article - 4%) occurred less frequently.

The structural disproportions (facial or corporal) were cited in 11 publications (44%)^{3,6-8,10-13,15-16,22}, that affect the type and facial proportion. The facial asymmetries were mentioned in five articles (20%)^{3,6,8,11,15} and the corporal ones in two (8%). Changes in cranial base^{6,7}, cephalic growth retardation¹⁶, the microcephaly^{12,13} or macrocrania²² and short face with disproportion between the middle and lower thirds of face⁸ were also cited.

Other disorder mentioned was oropharyngeal dysphagia with or without signs of aspiration, being related in sever researches (28%)¹⁴⁻²⁰.

The presence of persistent primitive reflexes or even pathologic were described in six publications^{2,15,17-20} (24%) associated to neuromotor disorders of the non-progressive encephalopathy (isolated)¹⁷⁻¹⁹, of the syndromic^{2,15} conditions or related to other diseases²⁰. The altered sensitivity

(intraoral or extra) were reported in four publications (16%)^{14,15,18,26}.

The abnormality in the muscle tone of neurological nature (of speech and corporal organs) was mentioned by the majority of the consulted literature^{2,3,6,8-10,13,15-18,20,22,26} (in 16 articles - 64%), being the hypotonia^{2,3,6,8-10,13,18,20,22,26} the most reported in literature (11 - 44%), distributed as follows: lips^{2,3,6,9,11,18,26} (7 - 28%), cheeks^{2,3,6,9,26} (5 - 20%), masseter muscles⁸ (1-4%) and temporal⁸ (1-4%). The tongue reduced tension was also described by literature^{2,8,9,11,12,26} (6-24%) and the body hypotonia in congenital myasthenic syndrome¹⁶ (one article - 4%). The spasticity was mentioned in one article (4%) related to non-progressive encephalopathy¹⁷ and rigidity in Sturge-Werber syndrome¹⁵ (one publication - 4%).

Several changes have been cited in the literature about the mobility of the speech articulator organs, such as hypomobility^{2,6,8,14,16,18,22,25,26} (in nine publications - 36%, being tongue^{3,6,14,18,26} and lips the most cited ones^{3,14,18,26}), the incoordination / inaccuracy of movements^{6,8,20} (three - 12%), restriction of mandibular movements^{14,15} (two - 8%) and the hypermobility / hyper-functionality^{8,25} (two articles - 8%). Changes in mobility were cited in 12 publications^{2,3,6,8,14-16,18,20,22,25,26} (48%).

Most publications^{3,6-8,10-15,18,20,22,23,26} reported speech disorders (15 - 60%) with developmental delays^{3,11-15,18,20} (including absence^{12,15} and restriction¹³ of the speech, eight articles - 32%), unintelligible^{3,8,10,11,14} (including inaccuracy⁸, five - 20%), dysarthria^{14,20} (two - 8%) lisp⁸ (1- 4%), increased speed³ (1 - 4%), an excess of salivation during speech⁷ (1 - 4%), phonetic-phonological disorder⁸ (1 - 4%) and resonant¹⁴ alterations (hypernasality with nasal air escape, 1 - 4%). Other studies^{6,7,22,23,26} (five - 20%) affirmed changes in speech without, however, describing it.

The changes in the breathing mode^{3,8,9,11,12,15,18,23} publications were cited in eight publications (32%), citing oral^{3,8,9,11,23} and oronasal^{2,15,18} breathing. Other respiratory disorders have been cited as apnea^{20,23} (8%), the presence of noisy breathing²⁰ (4%), inspiratory stridor²⁰ (4%), snoring²³ (4%) and bronchitis¹⁸ (4%).

From 11 (44%) researchers^{3,4,8-12,14,18,21} who evaluated chewing, all of them reported chewing alterations. The changes mentioned were: chewing with open lips (4 surveys^{3,4,9,12} - 16%), predominance

of vertical jaw movements (3-12%)^{3,10,12}, kneading the food with their tongue (3 - 12%)^{9,12,18}, changed speed (2)^{3,9}, inefficient / poor chewing (2-8%)^{4,10}, noisy (2-8%)^{4,12}, among other changes with a quote, the equivalent of 4% (unilateral³, change in food incision³, difficulty in lateralization of the food bolus¹⁰, mandibular deviation¹¹, exhaust food¹² and use of compensatory movements¹⁴). Three studies reported that there was no difficulty in this oral function, however, they describe the changes^{8,21,23} occurred.

Most of the raised studies (15 - 60%) cited dental abnormalities, including malocclusion (9 - 36%)^{2,3,6-8,10,13,15,19} the presence of periodontal disease (6 - 24%)^{4-8,19}, dental caries (4 - 16%)^{2,6,24,25}, changes in tooth (3 quotes regarding size^{6,10,16-12%}, one⁶ presenting shape alteration and another one in the dental enamel², each with 4%), poor hygiene (two - 8%)^{4,14} and delayed tooth eruption (two - 8%)^{2,6}.

Swallowing alterations were highlighted by most of the authors (20 researchers - 80%)^{2-5,8-21,23,25}. The hypersalivation in six (24%)^{12-14,18,19,25}, the presence of residue after swallowing in four (16%)^{3,11,12,14}, atypical or adapted swallowing in

three (12%)^{5,8,9}, with tongue interposition in three (12%)^{3,11,12}, with the presence of compensatory movements (3 - 12%)^{11,14,15}, food exhaust in two (8%)^{15,18}, open mouth swallowing (one - 4%)¹⁸, nasopharyngeal regurgitation (one - 4%)¹⁹, and seven articles (28%)^{2,4,10,13,19,21,23} there was a citation of difficulty, without, however, describing it. Regarding the oropharyngeal dysphagia, seven surveys cited it¹⁴⁻²⁰ (28%).

Orofacial myofunctional disorders in persons with ID, highlighted by the literature, in decreasing order of occurrence were: difficulties in swallowing (in 80% of selected texts)^{2-5,8-21,23,25}, changes in tonicity (64%)^{2,3,6,8-10,13,15-18,20,22,26} and structural (including dental - 60%)^{2,3,6-13,15-16,21,22,25} of the stomatognathic system, changes in posture^{3,6,8-13,15,18,19,23} and in the mobility of the phono-articulator organs^{2,3,6,8,14-16,18,20,22,25,26} (48% each), difficulties in chewing^{3,4,8-12,14,18,21,23} (44%) and respiratory changes^{3,8,9,11,12,15,18,23} (36%).

Table 2 briefly explains the different conditions showed that, according to the survey in literature, people with intellectual disabilities and orofacial myofunctional disorders²⁻²⁶.

Condition	Orofacial structures	Muscle tone and Mobility	Functions			
			Breathing	Chewing	Swallowing	Speech
Cornelia Lange Syndrome ²	Antiverted nostrils, "carp mouth" microstomia, thin upper lip; micrognathia, increased interocular distance; extended nasolabial furrow and antimongoloid palpebral fissures, narrow palate, delayed tooth eruption, enamel hypoplasia and atresia of the dental arches.	- Hypotonia (except in mentalis muscle) - Decreased mobility.	-	-	Difficulty in swallowing for solid and frequent choking.	-

Table 2 - Characteristics of orofacial myofunctional disorders associated with intellectual disability

Condition	Orofacial structures	Muscle tone and Mobility	Functions			
			Breathing	Chewing	Swallowing	Speech
Fragile X Syndrome ³	- Long face, mild facial asymmetry, parted lips at rest (with the possibility of sealing), high-arched palate, and malocclusion, anterior and in mouth floor tongue posture.	- Tongue and soft palate with good mobility.	Probable oral breathing mode with the clavicle rise in inspiration and little movement of the abdomen.	Unilateral chewing accelerated, with vertical movements, performed with open mouth and lateral incision.	Presence of hypersalivation, swallowing with tongue interposition and presence of food residue after swallowing.	-Unintelligible speech, restricted and accelerated.
Down Syndrome ⁴⁻⁹	Periodontal disease ^{4,6,7,8} , occlusal changes ⁴ , short face ^{6,8} ; underdevelopment of cranial base ^{6,7} ; (pseudo) macroglossia ⁶ ; fissured tongue, with teeth marks on its sides ⁶ ; parted lips (with the tongue between the lips) ⁶ ; dental abnormalities of eruption ⁶ , in shape (peg-shaped teeth) ⁶ number (agenesis and supernumerary teeth) ⁶ , size (microdontia) ⁶ and structure (with hypoplasia and hypocalcification) ⁶ ; narrow palate ⁶ or high-arched ⁹ ; small nose ⁶ ; malocclusion ^{6,7,8} ; hyperplasia of tonsils ⁶ and dry lips ⁶ .	Lips ^{6,9} , cheeks, temporal ⁸ and masseter ⁸ Hypotonia, tongue tension decreased ^{6,8,9} - Inaccurate and slow ⁶ tongue movement; hyper-functionality of the temporomandibular joint and hypomobility ⁸ of the phono-articulator organs ⁸ .	Oral ^{6,8,9} .	Difficulties in chewing ^{4,8} , with parted lips ^{4,9} , unilateral ⁴ , change in velocity ^{4,9} , noisy ⁴ , little strength in biting ⁴ , difficulties with consistent food ⁴ and kneading the food with their tongue ⁹ .	Swallowing difficulties ^{4,8} , atypical ^{5,9} or adapted and compensatory movements in its various phases ⁵ .	- Changes in speech ^{6,7} , salivation excess during speech ⁷ , articulatory inaccuracy, lisp, phonetic-phonological disorder ⁸ .

Continuation Table 2 - Characteristics of orofacial myofunctional disorders associated with intellectual disability

Condition	Orofacial structures	Muscle tone and Mobility	Functions			
			Breathing	Chewing	Swallowing	Speech
Silver-Russell Syndrome ¹⁰	Prominent occipito-temporal circumference, microdontia, micrognathia, triangular face, small ears, high-arched hard palate, furrowed tongue, body asymmetry, parted lips and tongue habitual posture on the floor of the mouth.	-Muscle hypotonicity.	-	Decreased grip strength in food and rotation movements and lateralization of the bolus.	Alteration of the oral phase of swallowing.	-Speech unintelligibility.
Goldenhar syndrome ¹¹	Facial asymmetry, deep hard palate, mixed dentition in good condition and asymmetry of the lower lip.	-Lips hypotonia and tongue tension decreased.	Oral.	Bilateral chewing, without centralizing the bolus and mandibular deviation.	Swallowing with tongue interposition, anteriorization of the head and food waste in vestibule after several swallows.	-Delay in speech development and affected intelligibility.
Angelman Syndrome ^{12, 13}	Microcephaly ^{12, 13} , parted lips habitual posture ¹² ; tongue protraction ^{12,13} , Symmetric cheeks ¹² ; Normal hard palate; dentition in good condition ¹² ; habit of opening and closing mandibular (without the presence of food, simulating mastication) ¹³ , prognathism ¹³ , wide mouth ¹³ , thin upper lip ¹³ , the presence of diastema and primate spaces in the dental arches ¹³ .	- Decreased tongue ² tension and muscular ¹³ hypotonia.	Mixed (predominantly oral) ¹² .	Chewing with mouth opened ¹² , with vertical movements ¹² , kneading food with the help of the tongue ¹² , presence of constant noise ¹² and food scape during chewing ¹² .	Tongue interposition when swallowing (without coughing or choking) ^{12,13} , hypersalivation and presence of food residues in the mouth ¹² .	- Absence of functional speech ^{12,13} , producing 2 to 5 syllables and 3 to 4 words ¹³ .

Continuation Table 2 - Characteristics of orofacial myofunctional disorders associated with intellectual disability

Condition	Orofacial structures	Muscle tone and Mobility	Functions			
			Breathing	Chewing	Swallowing	Speech
Worster-Drought Syndrome ¹⁴	Poor oral hygiene and lack of lip sealing.	Hypomobility of the lips, tongue and jaw, bucofacial dyspraxia.	-	Preference for foods with lower consistency and compensatory movements.	Neonatal dysphagia, gastro-esophageal reflux; presence of hypersalivation, Food residues and compensatory movements.	- Delay in speech development, most with unintelligible speech, presence of dysarthria, hypernasality and nasal air escape
Sturge-Werber syndrome ¹⁵	Postural (shoulders, head, jaw, parted lips and tongue) and craniofacial (asymmetry of eyes, nose, ears, lips, cheeks, tongue, hard and soft palates, dental midline, mentalis muscle and possible jaw discrepancy) changes, malocclusion, alterations in sensibility, presence of primitive searching and sucking reflexes.	- hypertonic lips - Restriction of mandibular mobility.	Oronasal.	-	Hypersalivation, swallowing and head movements involving periorbicular muscles	- Absence of orality and / or lack of patient cooperation
Congenital myasthenic syndrome ¹⁶	Microretrognathia, high-arched palate and small palpebral fissures, insensitivity to pain, with self-injurious behaviors (such mutilation of the tongue tip), teeth anomalies (central incisors with the size of the permanent dentition) and delay of head growth.	Body hypotonia - Hypomimia (reduction and slowness of facial movements in the execution of facial movements).	-	-	Dysphagia	-

Continuation Table 2 - Characteristics of orofacial myofunctional disorders associated with intellectual disability

Condition	Orofacial structures	Muscle tone and Mobility	Functions			
			Breathing	Chewing	Swallowing	Speech
Nonprogressive encephalopathies ¹⁷⁻¹⁹	- Presence of pathological reflexes ^{17,18} , difficulty in lips sealing ¹⁷⁻¹⁹ , hypofunctioning and shortened upper lip and the lower with eversion ¹⁸ ; asymmetric cheeks ¹⁸ , jaw with slight deviation to the left side ¹⁸ , interposed tongue at rest ¹⁸ , reduced intraoral sensitivity ¹⁸ changes in posture of the tongue ¹⁹ and in dental tissues, dental ¹⁹ malocclusion (overbite overject and crossbite), periodontal diseases ¹⁹ and gingival hyperplasia ¹⁹	- Hypotonic lips and cheeks ¹⁸ - Reduced mobility of the jaw and the tongue ¹⁸ .	Oronasal breathing mode and with predominant movement of the abdomen ¹⁸ .	Kneading the food with the tongue against the hard palate ¹⁸ .	Dysphagia ^{17,19} , fluid escape through the labial commissures ¹⁸ and hypersalivation ¹⁹	- Changes in speech development ¹⁸ .
Lesch-Nyan Disease ²⁰	Hyperreflexia and dystonia (including the tongue).	- Generalized hypotonia in most cases. Some patients had spasticity and rigidity.	Respiratory abnormalities (inspiratory stridor, apnea, cyanosis, and noisy breathing).	-	Disphagya.	- Delayed speech development and presence of dysarthria.
Neurofibromatosis type ^{1,21,22}	Oral neurofibroma located in the hard palate (hard palate's volume increased) ²¹ , extending of the space of the mandibular canals ²² , resorption of alveolar bone crests ^{21,22} , body asymmetry and macrocrania ²² .	-Reduced muscular strength ²² , velopharyngeal and oropharyngeal muscles insufficiency (hypotonia) ²² - Lowered or regular ²² mobility of the soft palate.	-	Complaining about difficulty in chewing ²¹ .	Complaining about difficulty in swallowing ²¹ .	- Speech disorders ²² .

Continuation Table 2 - Characteristics of orofacial myofunctional disorders associated with intellectual disability

Condition	Orofacial structures	Muscle tone and Mobility	Functions			
			Breathing	Chewing	Swallowing	Speech
Mucopolysaccharidosis ²³	Dental arch and tongue commitment.	-	Apnea, mouth breathing more frequent in the group without enzyme replacement therapy	Changed	Escape of saliva during sleep and swallowing disorders.	- Speech disorders.
ID alone (or without description) ²⁴⁻²⁶	Cariogenic disease ²⁴ and macro and microglossia ²⁵ .	- Decreased tongue tension ²⁶ , cheeks and lips hypotonia ²⁶ .	-	-	Hypersalivation ²⁵ .	Speech disorders ²⁶ .

Continuation Table 2 - Characteristics of orofacial myofunctional disorders associated with intellectual disability

Most studies did not mention the level of ID, being recorded in five articles^{2,10-12,16} (20%), being considered light in one article¹¹ (8%), mild to moderate in one¹⁰ (4%) moderated in one¹⁶ (4%) and severe in two^{2,12} (8%).

Discussion

Restrictions on people's daily lives with ID may be different, affecting everything from learning to social activities, depending on different factors such as the seriousness of this deficiency, the environment¹, the state of health, of comorbidity factors associated or not to the condition, and the different syndromes²⁻¹⁶ or disorders¹⁶⁻²³ that may coexist, besides the need for specialized professionals in the area and teams that operate in an interdisciplinary way, in order to offer a better quality of life and health of these individuals and

their families. There are even situations where the diagnosis is delayed or the etiologic factor of the deficiency²⁴⁻²⁶ is not known, hindering the implementation of preventive measures in the area.

The proposal of a systematic review study took place in order to verify the scientific evidence about the topic of this research proposal, it should be emphasized that among the descriptors used, the term "oral motricity" was selected and not "orofacial motricity" due to the inclusion of the period in which the nomenclature was still this (after CFFa Resolution N° 320, of February 17, 2006, the area's name was changed to orofacial motricity).

Regarding to the period when search was performed, there was a tendency for increasing the number of researches about ID in OM area, with the highest concentration of publications occurring in 2010. The writings of the speech therapist have increased considerably, and in the 1990's there

was a great increase of publications in OM area in scientific articles, most probably by the supply and demand for professional training in specialized courses in the area, especially in relation to the creation of the CEFAC; the creation of stricto sensu courses and a CAPES requirement for a better qualification in such courses, to the increasing number of graduation courses in Speech Therapy and the creation and organization of journals that circulated specific area articles²⁸. From 1990 to 2012 there was a sharing of information available through the Internet and an increasing of lato sensu courses supply, which may explain the increase in scientific production in a continuously way.

Regarding the type of scientific production, there was not a dominance of quantitative (40%) or qualitative (36%) researches and, in a lower proportion, the publication of theoretical research^{6-8,13,19,25} (24%). Regarding the non-prevalence of quantitative or qualitative research, the results obtained show that researchers in this area are much deeper into the subject, and are aware of the most prevalent characteristics in these individuals. Most of the articles presented descriptive study (76%), this fact can provide a good quantification of the scientific study that will be analyzed.

In relation to theoretical productions, which in hierarchical order are those with greater scientific evidence base, no satisfactory results were obtained, since studies containing meta-analysis, review studies with inclusion of cohort studies and clinical trial with Control Group were not obtained, showing that from six theoretical articles, four^{6,7,19,25} were critical review or narratives and only two articles^{8,13} were integrative review (Table 1). These results may lead to the assumption that the systematic review of the researches needs more time for the researcher to a more attentive and careful reading on a particular subject, it may generate less interest in conducting this type of research, although more effort is necessary from the researchers, in order to produce publications with the higher levels of scientific evidence.

It is noteworthy that two clinical trials (longitudinal studies involving therapeutic process)^{12,18} were included in this review, being used evaluation data, so that there would be no interference of the phonotherapeutic intervention in the description of orofacial myofunctional characteristics (Table 1). The clinical trials showed good level of scientific

evidence when are used Control Groups, they are randomized and the groups are homogeneous, which could not be seen in obtained publications.

In relation to the cross-sectional studies, the projects which present the greater scientific evidences are those consisted of homogeneous groups, with control and definition of small confidence intervals in the statistical treatments. Accordingly, in ten publications^{4-5,9,14,17,20,22-24,26} (40%) taken as such studies (Table 1), it may be noted heterogeneity in the age of the individuals that constituted the study samples^{4,5,9,14,17,20,22-24}, except three, in which a publication did not mention age²⁶, other mentioned just the average age¹⁴, and another cited that the participants were aged up to four years old⁹. The groups were homogeneous regarding the investigation of the same disease in six of the transversal studies^{9,14,17,20,22,23}, being important to emphasize that even then, in some situations there may be conditions with different motor changes (as in Lesch Nyhan syndrome, in which the entire sample had motor impairment but with different manifestations, depending on the lesion location²⁰) or presence of different subtypes of the same disorder (for example, in mucopolysaccharidosis and severity of their manifestations²³). The presence of a control group was reported in the discussion of a single article²² (although there was not a description of the method) and the statistical treatment was reported in four studies^{14,22-24}, although only in three^{14,22,24} there was a description of the range of confidence. Thus, it can be stated that there is a need to establish better criteria in the composition of samples for cross-sectional studies, so they can offer, in relation to orofacial myofunctional characteristics, scientific evidence with higher quality.

It is noteworthy that in the case and cross studies, a variable that deserves distinction relates to the diversity of the age group of the studies, because the age range of study comprised individuals aged between six months and 67 years old, although 64% of studies involving humans showed, mostly, the childhood (from zero to 12 years old).

It is known that the functions of the stomatognathic system are performed differently throughout life because of the socio-cultural and environmental differences, the craniofacial growth itself, the change in eating habits, the eruption and replacement of teeth and maturation of the central nervous system to control and coordinate each

function operation. Moreover, it still has to be considered the aging of structures, the structural change (teeth, mucosa, bone, temporomandibular joint, salivary glands and others), of the sensory perception (smell, taste and stomatognosy) and functional (orofacial muscles with lower strength and coordination of movements)²⁷.

It is hypothesized that some researchers^{4,5,17,20,22-24} have conducted studies with different ages due to the difficulty in obtaining data that depend on the cooperation of patients with ID¹². Furthermore, in research involving human beings the failure to attend the schedules intended for the search procedure may occur for several reasons, such as financial, lack of interest or even the fragility of the health condition of these individuals. The difficulty in understanding what is proposed in exams is also a possibility, and this variant is cited by literature⁴.

Regarding the gender, of the articles that were presented as clinical reports and cross-sectional studies, the prevalence was higher in males (395 cases - 59.94%) than in females^{2,4-5,14,16-18,20-24} (264 - 40.06%). These results contradict the data of the IBGE census of 2010²⁹, in which the females have more disabilities (listening, visual, motor and intellectual) than males. This discrepancy may have occurred for several reasons, among which the Census is based on complaints from the population, while the consulted studies were the evaluation and diagnosis of specialists. Another factor may arise from the inclusion of ID with other disabilities, and in this case, there is a greater frequency of cases in men, but as this deficiency is included in a single denominator, i.e. "disability", there may be a bias in this data. Furthermore, it is hypothesized that the higher incidence in male subjects occurred due to the high association between learning disability and genetic and hereditary factors. As it has been previously stated, the authors²³ described the presence of different mucopolysaccharidosis, which are resulting from genetic and hereditary enzyme deficiency disease in 56 individuals (from 78 in a sample).

Most studies (84%) involved evaluation or diagnosis (doctor or speech therapist)^{2-11,13-17,20-24,26}, due to a higher occurrence of cross-observational and descriptive studies. The treatment was described in 28% of the studies^{7-8,12,18-19,24-25} (it is

important to note that in three studies, there was also the description of the evaluation^{7-8,24}). So, it is highlighted that longitudinal studies require more time for scientific research, which can cause some difficulties in scientific research, such as the abandonment of therapies, excessive absences or other variables that may compromise the results. This tendency in the thematic of evaluation and diagnosis was also observed in the literature²⁸, in the period from 1970 to 2000.

In practical clinic (speech therapy and dental), some ID patients do not cooperate with opening the mouth, do not accept touch, do not understand the orders given by the professional and have sudden behavior change, disfavoring a more detailed evaluation and, therefore, damaging the treatment's results, as well as results collected in a less reliable evaluation process. The mentioned considerations were ratified by the literature²³, reporting that in some situations, the evaluation of the stomatognathic system is not completed or even effected due to the deficits shown by patients (cognitive, joint and visual).

Only in four studies^{2,4,10,19} (16%) the description was interdisciplinary, showing that health practices still need further rapprochement between the different areas. The performance and interdisciplinary research also require, from the involved professionals, the study of the related areas and more time available for group meetings, which is not always possible or interesting for those involved. It is noteworthy that most of the studies (60%) involve the presence of syndromic conditions²⁻¹⁶, requiring interdisciplinary approach, which did not result in publications with the stamp on interdisciplinarity, unfortunately.

Therefore it can be inferred that the discussion / conducting of the researches in an interdisciplinary manner in the area of ID, as well as clinical practices involving rehabilitation still need further strengthening on the part of speech therapy and other Health areas.

The research involving OM in individuals with ID were characterized, with higher prevalence, by syndromic conditions (56%), evidencing the interest of researchers in genetic changes. Despite this, with the exception of Down syndrome, other syndromes have few phonoaudiological studies

reporting orofacial myofunctional characteristics, suggesting further research in the area.

Despite the heterogeneity of the studies, some considerations regarding the results obtained about the orofacial myofunctional aspects of people with ID will be held.

The structural changes were described by most researchers (60%), related or not to syndromes^{2,3,6-13,15,16,19,22,25}, whereas the change in size, shape or volume of the hard palate^{2,3,6,9,11,16,21}, the maxillo-mandibular discrepancies^{2,8,15,16,21} and asymmetries (body and facial) were the most frequent findings and it is known that structural alterations of the stomatognathic system undermine and limit the performance of the functions of this system, requiring the actions of other health areas such as Orthodontics, Functional Orthodontics, Orthopaedics and correction of bone structural aspects, so that, the Speech Therapy can act.

Although changes in posture (of phono-articulator and body organs)^{3,6,8-13,15,18,19,23} are not cited by most articles, they affect breathing, chewing, swallowing, speech and occlusion, especially the altered position of the tongue^{3,6,8,9,10,12,13,15,18,23} and lips^{3,6,9,10,11,12,15,18,19} which were the most cited changes in the articles. It is noteworthy that the position of the phono-articulator organs depends on the interplay of a numerous factors, such as bone condition, the tone of the orofacial muscles, musculoskeletal and structural harmony of the stomatognathic system and good living and health conditions. Through the consulted literature, it can be seen that many of the conditions cited for a postural adaptation were impaired in ID, because there is often the presence of hypotonia (lips^{2,3,6,9,11,18,26} and cheeks^{2,3,6,9,26}, reduced tongue tension^{2,8,9,11,12,26} and body¹⁶); structural disharmony of the stomatognathic system^{2,3,6-11,13,15,16,19,21,25} (such as micro²⁵ or (pseudo) macroglossia^{6,25}, shape, size and volume of the hard palate^{2,3,6,9,11,16,21} and maxillo-mandibular disproportions^{2,8,15,16,21}, which lead to occlusal changes^{2,3,6-8,10,13,15,19}) and bad respiratory tract conditions^{18,20,23} (as bronchitis¹⁸, inspiratory stridor²⁰ and apneas^{20,23}). Therefore, it must be reflected, in clinical practice, the current conditions of such individuals when planning the work for the appropriate positioning of the body and of those structures.

Other aspects that deserve attention, still in relation to structural characteristics and that affect directly in the posture and execution of orofacial functions relate to the conditions of cephalic growth, to the facial type and proportion, usually disproportional in subjects with ID, as can be seen in literature^{3,6-8,10-13,15-16,22}. Therefore, the unfavorable prognosis for the stomatognathic system adequacy in individuals with this type of disability should be considered, because of the structural conditions above.

The abnormality in the phono-articulator and body organs tone was cited by 64% of the consulted literature^{2,3,6,8-10,13,15-18,20,22,26}, with hypotonia^{2,3,6,8-10,13,18,20,22,26} being cited more than spasticity¹⁷ and rigidity¹⁵. It is known that there may be bone changes resulting from changes related to the muscle tone, which may hamper the independence / autonomy and the execution of the stomatognathic system functions. The altered muscle tone also impairs the proprioceptive function, which provides information about the condition of the muscles and tendons as well to their status and movement - providing adjustments in pressure and muscle tension necessary for static or dynamic⁸ attitudes. The literature^{6,7} also highlighted that, associated with generalized hypotonia in Down syndrome, the temporomandibular joint dysfunction^{6,7} and the hiperlaxitude⁶ of movements may occur, which can promote the emergence of bruxism, tooth wear and future tooth fracture⁶. Furthermore, it is known that body hypotonia hinders a proper posture, causing fatigue in a shorter time than in individuals with suitable muscle tone. The same occurs in hypotonia of the phono-articulator organs, generating, according to the literature⁷, difficulties in performing the functions of the stomatognathic system, especially in Down syndrome, in which the hypotonia is widespread - this aspect, in particular, difficults the maintaining of the results in orthodontic treatments, in which the final phase of the treatment (containment), the muscle exerts great influence on the occlusal stability⁷. Therefore, early interventions to minimize the effects of abnormalities related to the muscle tone are recommended.

The changes, in 48% of the cases, in the mobility of the phono-articulator organs were described by the consulted researchers, being described the

hipomobility^{2,6,8,14,16,18,22,25,26}, the incoordination / inaccuracy of movements^{6,8,20}, the restriction of jaw movements^{14,15} and hypermobility / hyper-functionality^{8,25}). The altered and performed in an imprecise and uncoordinated way mobility, linked to structural, sensitive and proprioception changes can lead to difficulties in sucking⁸, swallowing^{2,8,14}, in synchronizing functions as chewing, swallowing and breathing, and can generate dysphagia^{2,14}, in speech production⁸, so that, parameters as speed, accuracy and rhythm can be guaranteed; in facial expression^{14,16}, in addition to difficulties in oral hygiene^{14,25} and feeding¹⁴.

The speech neurogenic disorders, registered in most of the consulted articles^{3,6-8,10-15,18,20,22-23,26} (60%) can compromise significantly the life of individuals with ID, since this motor act allows the “Other Person” to understand the message expressed, being possible to happen impairments of social interactions¹⁴, especially when the condition comes accompanied by changes in language, common because of cognitive impairment. In some syndromes, such as Angelman^{12,13} and Sturge-Werber¹⁵, may be provided from absence¹⁵ to speech restriction¹³, and it is often necessary to use alternative communication. Furthermore, speech disorders may be explained by the loss in mobility, muscle tone, sensitivity and proprioception²⁶; by structural changes, particularly in Down syndrome by macroglossia⁷; by the delay in the development of these subjects that, concomitantly, have speech development delay^{3,11-15,18,20}. The lack of coordination of orofacial muscle groups^{6,8,20}, the restricted mobility^{2,6,8,14-16,18,22,25,26}, the change in body and phono-articulator organs tone^{2,3,6,8-10,13,15-18,20,22,26} as well as a decreased tactile orofacial sensitivity²⁶ can provide speech unintelligibility^{3,8,10-11,14} described in the consulted publications. Other aspects that can also impair speech are changes in breathing^{3,8,9,11,12,15,18,23} and resonance¹⁴ cited in 36% and 4% by the consulted articles respectively. The dysarthrias^{14,20} were present, showing the involvement of motor neurological basis for speech. The diversity of the methods of the studies and the different objectives of the researches consulted were variables that prevented a real analysis of speech disorders of individuals with ID, it is important to emphasize that the focus of this research did not include analysis of such changes, but because of the selected articles mention such impairments, the

results were added. Review studies including the descriptors: “speech”, “disorders of articulation”, “pathology of speech and language” and “speech disorders” would be important to a greater extent about the subject.

Mouth breathing^{3,8,9,11,23}, oronasal breathing^{12,15,18}, the apnea^{20,23}, bronchitis¹⁸, noisy breathing (inspiratory stridor)²⁰ and snoring²³ were cited by the selected publications. It is known that when the breath is not performed by the nose, it can cause different consequences, usually associated with events such as malocclusion, speech disorder, postural changes in craniofacial development, in execution of oral functions (such as chewing and swallowing) among others. Although apnea and bronchitis are conditions less cited by the authors consulted, they deserve special care so that other events do not undermine even more the ID individuals’ life and health. Thus, the conditions that prevent nasal breathing should be investigated, being important to establish partnership with Otorhinolaryngology.

Chewing was seen as altered in 44% of the articles, showing patterns of immaturity in chewing pattern by virtue of the neuroanatomophysiological aspects in ID. Another factor to consider that focuses on chewing difficulty relates to the oral health of people with ID. The lack of manual dexterity, the difficulty in understanding the importance of dental hygiene, the high prevalence of dental caries, periodontal disease, poor dental formation (including agenesis, teeth changes in size and shape) and malocclusion aspects are to be considered as aggravating for an efficient and harmonious chewing.

Confirming the foregoing, the conditions related to oral health^{2,4-8,10,14,16,19,24,25}, changes of eruption delay^{2,6}, in size^{6,10,16}, shape⁶ and enamel² of teeth and malocclusion^{2,3,6-8,10,13,15,19} were alterations cited by most authors consulted, requiring an interdisciplinary approach with Dentistry and Orthodontics. Also, the performance with the family, emphasizing the importance of oral hygiene and of monitoring, from an early age, with dentists, so, the condition of oral health does not harm the living conditions and the stomatognathic functions seem to have a fundamental importance for individuals with ID.

Swallowing alterations were highlighted by most authors (20 researchers - 80%)^{2-5,8-21,23,25}, and adaptations are necessary in some situations

because of craniofacial structural abnormalities. It is worth noting that the process of swallowing is a complex act that requires integration with other stomatognathic functions (such as breathing and chewing), with sensitivity (laryngeal and extra and intraoral), with the synchrony the contraction of muscle groups involving the mobility of the tongue and jaw, with the occlusive conditions, so that, there will be a mandibular stability, with the muscle tone, so there will be enough intraoral pressure and with neurological integrity, which is involved in all phases of swallowing. These conditions, taken by the majority of the studies as amended, greatly interfere in swallowing and in the health of individuals with ID.

Dysphagia (reported in 28% of searches)¹⁴⁻²⁰ and difficulty in swallowing saliva (24%)^{12-14,18,19,25} are conditions that can lead to bronchoaspiration, aspiration pneumonia and malnutrition, which can harm the health conditions, and also can to administration of alternative feeding route, compromising even more the feeding difficulties of these individuals. Such demonstrations require services in specialized and interdisciplinary health as early as possible, since its detection to its restoration (when possible) of the safe swallowing process.

In reporting a clinical case of an individuals with ID and Sturge-Weber syndrome, the inappropriate positioning of the tongue, the morphofunctional changes of the phono-articulator organs and the inability to control the laryngeal sphincter, characterize the dysphagia condition found, since the lack synchrony between elevation and anteriorization of the larynx and glottal closure, and failure in the displacement of the epiglottis over the laryngeal aditus as the mechanism of the protection of the lower respiratory tract by apnea were factors that hindered safe swallowing in the presented case¹⁵.

The presence of persistent primitive or even pathological reflexes were reported in 24% of selected articles associated with neuromotor dysfunctions of nonprogressive encephalopathy (isolated)¹⁷⁻¹⁹, of the syndromic conditions^{2,15} or related to other diseases²⁰. The altered sensitivity (extra or intraoral) was reported in 16% of the publications^{14,15,18,26}. The immaturity of the central nervous system hinders the autonomy to perform functions such as chewing and swallowing accurately, coordinated and volitional, also affecting the

growth and the weight and height development, the individual's health, requiring the performance of multidisciplinary teams and family support for the act of feeding, which often cause modifications in the routine of daily life for everyone involved. There is also the food consistency change, usually for more pasty, which will tend to make the orofacial muscle tone flaccid, but it will result in a safer feeding, since with the presence of pathological reflexes it is associated with neurogenic dysphagia, as mentioned in the literature¹⁷, which may, by virtue of the aspirations, compromise the lower respiratory tract¹⁹. The literature also commented on the difficulty of containing the saliva in the intraoral cavity, which may result in a necessity to the responsible in always having at their disposal various bibs, and it also can cause social embarrassment during the act of feeding¹⁸. The presence of pathological reflexes, as biting, can cause difficulties in cleaning the oral cavity, tooth wear and promote the appearance of bruxism, leading to the emergence of periodontal and occlusal changes¹⁹. Thus, orofacial myofunctional losses may hinder the feeding and quality of life of individuals with cognitive damages⁵.

Another aspect that deserves attention in the act of swallowing is the body posture, especially the head, as the cervical hyperextension evident in some cases of non-progressive encephalopathies (NPE), complicating the fixation of the hyoid bone to the laryngeal elevation required for closure of the epiglottis and protection of the lower respiratory tract¹⁷.

Besides the presence of pathological reflexes, previously mentioned, the increased intraoral sensitivity in NPE were aspects that hampered swallowing, according to the literature¹⁸. The change of muscle tone²⁰ was also one of the reasons cited to explain the difficulty in implementing this oral function.

The difficulties in feeding, mainly on the evidence of neurogenic oropharyngeal dysphagia, bring serious consequences for the health of the individual, requiring assistance from a responsible during feeding, especially when the responsible is aware of grievance factors and risk of dysphagias, so that, the responsible can proceed with proper care in this process (since the preparation of food, supply, utensils, care for the individual's

environment, the pace of supply and possible essential maneuvers to a safe swallowing, depending on each situation, even patience and availability of time). Additionally, there may be food refusal because of the perceived difficulties by the person with ID (as cough and frequent choking), turning the act of feeding in a time full of aversive behaviors, avoiding also foods with certain textures, as quoted in the literature¹⁴. Thus, the interdisciplinary approach is essential to minimize the risks and ensure a safer and more enjoyable food supply.

Most studies did not mention the level of ID, being recorded in five articles (20%)^{2,10,11,12,16} from these it was considered as mild in one article¹¹, mild to moderate in other publication¹⁰, moderate¹⁶ in another one (4%) and severe in two^{2,12}. In this sense, the aspect of age at diagnosis of intellectual disability is emphasized, because generally the cases taken as mild present later diagnosis in contrast to the serious ones, which usually occur (with some exceptions) before the age of seven³⁰. It would be interesting to mention and distinguish the level of ID, because patients with mild difficulties have better cognitive conditions than those with severe ID and, therefore, tend to understand and collaborate more in the realization of the proposed procedures.

Regarding the retrospective studies that include analysis of records, due to the establishment of data collection, many professionals or Health students can file procedures and examinations and, so, if there is no standardization and if the files do not go astray, there may be, in the medical records, a lack of a more detailed patient data. Furthermore, many ID diagnoses do not describe the level of disability, making these studies with these individuals hard to classify. There is also the difficulty of obtaining the diagnosis itself. Usually parents search for several experts and when those have an underprivileged socioeconomic status, the lack of access and the high demand for public services hamper an early and accurate diagnosis.

Thus, it can be seen that the orofacial myofunctional disorders in people with ID, highlighted by the literature, as in swallowing, in muscle tone, in structural conditions of the stomatognathic system, in posture and mobility of the phono-articulator organs, in chewing and in breathing justify the action in OM, being important to note that the heterogeneity of the methods used by the consulted

authors and the co-occurrence of ID in different conditions are factors that do not allow to define unique features, so, the clinician must take into account these aspects. However, it can be stated that the performance in OM and in dysphagia with partnership with other professionals (Dentists, Nutritionists, Physiotherapists, Pulmonologists, Occupational Therapists, Pediatricians, Otorhinolaryngologists, etc.) are important and fundamental to an appropriate intervention to minimize the signs and symptoms exhibited by these individuals.

From the literature review, it can be seen that there is a great diversity of clinical features and factors that may present the associated ID, which complicates the interpretation of the results obtained in a homogeneous way. Additionally, the different research methods adopted by the researchers were also a factor that hindered the analysis of orofacial myofunctional characteristics of people with such disability. Therefore, further studies should be performed because there are a considerably large number of individuals with a diagnosis of intellectual disability in our country and understanding the functioning of the stomatognathic system in these individuals is important to offer them and their families a better life condition. Thus, it can be stated that the studies in this area are the key to a better understanding of neuroanatomophysiological aspects involved in the implementation of the stomatognathic function in people with this disability.

Conclusion

Based on the results, the characteristics of the scientific production within the period specified, it can be concluded that:

- There was a prevalence of clinical studies (cross - observational and case reports);
- The largest concentration of publications occurred in 2010;
- Most of the researches relating OM with ID were performed by Brazilian researchers;
- Regarding age, the studies comprised individuals with six months to 67 years old, with a greater focus on the age group of zero to 12 years, with prevalence of the male gender;

- The intervention type most used and described by the studies was diagnostic and evaluation (doctor or speech therapist);
- The syndromic conditions were the most described and
- Most researches did not mention the level of ID.

In relation to orofacial myofunctional characteristics, all articles cited orofacial changes, highlighting the difficulties in swallowing, changes in muscle tone, in the structures of the stomatognathic system, in the positioning of the tongue and lips, in the mobility of the phono-articulator organs, the inefficient chewing and impairment in breathing mode. However, the heterogeneity of the methods used by the consulted authors and the co-occurrence of ID in different conditions are factors that do not allow defining unique features of orofacial myofunctional disorders in people with ID.

References

1. Amiralian MLT, Pinto EB, Ghirardi MIG, Lichtig I, Masini EFS, Pasqualin L. Conceituando deficiência. *Rev. Saúde Pública* 2000;34(1):97-103.
2. Santos MTBR, Fernandes AS, Biziak TR. Síndrome Cornélica de Lange: enfoque interdisciplinar. *J Bras Odontopediatria Odontol Bebê*. 2001;4(21):419-23.
3. Barbosa AD, Muller MP. Síndrome do X frágil: alteração miofuncional e de linguagem. *Rev CEFAC*. 2003;5:309-12.
4. Almeida LG, Sandrim SA, Costa JA, Almeida SO, Chiappetta ALML. Alterações clínicas periodontais e mastigatórias em indivíduo com deficiência mental. *Rev CEFAC*. 2004;6(1):40-3.
5. Pilinová A, Matejcková E, Lencová E, Foltinová J, Pisa J. Orofacial dysfunctions, drinking regimen and quality of life – long-term prospective study. *Cent Eur J Publ Health*. 2006;14(4):189-92.
6. Macho VMP, Seabra M, Pinto A, Soares D, Andrade C. Alterações craniofaciais e particularidades orais na trissomia 21. *Acta Pediátrica Portuguesa*. 2008;39(5):190-4.
7. Roy RA. Ortodontia em pacientes com síndrome de Down [monografia]. Canoas(Rio Grande do Sul): Instituto de Ciências da Saúde FUNORTE / SOEBRÁS; 2008.
8. Barata LF, Branco A. Os distúrbios fonarticulatórios na síndrome de Down e a intervenção precoce. *Rev. CEFAC*. 2010;12(1):134-9.
9. Oliveira FF; Andran CMA, Ghirello-Pires CSA. Amamentação, hábitos orais deletérios e alterações fonoaudiológicas: um estudo sobre suas relações. V Mostra Interna de Trabalhos de Iniciação Científica, 26 a 29 de outubro de 2010, Centro Universitário de Maringá, Maringá.
10. Rossi NF, Ueda KH, Richieri-Costa A, Giacheti CM. Síndrome de Silver-Russel: relato de caso. *Rev CEFAC*. 2006;8(4):548-56.
11. Lima FT, Araújo CB, Sousa EC, Chiari BM. Alterações fonoaudiológicas presentes em um caso de síndrome de Goldenhar. *Rev. Soc Bras. Fonoaudiol*. 2007;12(2):141-5.
12. Eskelsen MW, Pacheco FB, Montibeller CG, Blasi HF, Fleig R. Introdução e desenvolvimento do uso da comunicação alternativa síndrome de Angelman: estudo de caso. *Rev. CEFAC*. 2009;11(2):228-36.
13. Maris AF, Trott A. A patogênese genética e molecular da síndrome de Angelman *J Bras Psiquiatr*. 2011;60(4):321-30.
14. Clark M; Harris R; Jolleff N; Price K; Neville BG. Wroster-Drought syndrome: poorly recognized despite severe and persistent difficulties with feeding and speech. *Dev Med Child Neurol* 2010;52(1):27-32.
15. Santos SVP, Cavalheiro LG. Síndrome de Surge-Weber: relato de caso dos achados da avaliação fonoaudiológica. *Rev. CEFAC*. 2010;12(1):161-70.
16. Raspall-Chaure M, Toro-Riera MD, Gratacós M, Cuenca-León E, Ferrer I, Indo Y, Roig-Quilis M, Macaya-Ruiz A. Insensibilidad congénita al dolor con anhidrosis asociada a síndrome miasténico congénito. *Rev. Neurol*. 2005;41(4):218-22.
17. Andregueti SVC, Furkim AM. O estudo videofluoroscópico da aspiração traqueal em pacientes portadores de paralisia cerebral do tipo quadriparésia espástica com queixa de deglutição. *Rev. CEFAC*. 2003;5:143-8.
18. Rosa CMR, Lopes AR, Santos FF, Motta AR. A crioterapia como recurso para diminuir a sialorréia em criança com disfunção neuromotora: relato de caso. *Rev. CEFAC*. 2005;7(3):300-6.
19. Santos ATL, Couto GBL. Atendimento odontológico ao paciente portador de paralisia cerebral. *Int J Dent*. 2008;7(2):133-41.
20. Jinnah HA, Visser JE, Harris JC, Verdu A, Larovere L, Ceballos-Picot I, et al. Delineation of the motor disorder of Lesch-Nyhan disease. *Brain*. 2006;129(Pt 5):1201-17.
21. Ribeiro BF, Pacheco DFS, Freitas RA, Souza LB, Miguel MCC. Neurofibroma oral associado à neurofibromatose tipo 1: relato de caso. *Odontologia. Clin.-Cientif*. 2008;7(2):157-60.
22. Souza JF, Toledo LL, Ferreira MCM, Rodrigues LOC, Rezende NA. Neurofibromatose tipo 1: mais comum e grave do que se imagina. *Rev Assoc. Med. Bras*. 2009;55(4):394-9.
23. Turra GS, Schwartz IVD. Avaliação da motricidade orofacial em pacientes com mucopolissacaridose: um estudo transversal. *Journal de Pediatria* 2009;85(3):254-60.
24. Pereira LM, Mardero E, Ferreira SH, Kramer PF, Cogo RB. Atenção odontológica em pacientes com deficiências: a experiência do curso de Odontologia da ULBRA Canoas/RS. *Stomatós*. 2010;16(31):92-9.
25. Oliveira ALBM, Giro EMA. Importância da abordagem precoce no tratamento odontológico de pacientes com necessidades especiais. *Odonto*. 2011;19(38):45-51.
26. Tavares AMS, Bonfim ACQ, Albuquerque GC. Alterações miofuncionais orofaciais que comprometem a fala em pacientes com deficiência intelectual atendidos no Centro de Integração Psicossocial do Ceará. *Anais do IV Encontro Brasileiro de Motricidade Orofacial*; 2011 jun. 10-1; Natal: Universidade Federal do Rio Grande do Norte; 2011.
27. Amaral AKFJ. Interface da motricidade orofacial com a gerontologia. In: Pernambuco LA, Silva HJ, Souza LBR, Magalhães Jr, HV, Cavalcanti RVA (Org.). *Atualidades em motricidade orofacial*. Rio de Janeiro: Revinter 2012; p. 123-33.



28. Alves MRM. A produção fonoaudiológica nacional em motricidade oral 1970-2010 [dissertação]. Curitiba (Paraná): Universidade Tuiuti do Paraná; 2002.
29. Instituto Brasileiro de Geografia e Estatística. Censo demográfico 2010: características gerais da população, religião e pessoas com deficiência. Brasília: IBGE, 2010.
30. Jelliffe-Pawlowski LL, Shaw GM, Nelson V, Harris JA. Risk of mental retardation among children born with birth defects. *Arch. Pediatr. Adolesc. Med.* 2003;157:545-50.