Feeding, Swallowing, and Chewing Disorders in Children with Down Syndrome: an Exploratory Review *

Alteraciones en la alimentación, la deglución y la masticación en niños con síndrome de Down: revisión exploratoria

Distúrbios de alimentação, deglutição e mastigação em crianças com síndrome de Down: uma revisão exploratória

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ABSTRACT

Background: Down syndrome is the most prevalent genetic disorder associated with intellectual disability. It is characterized by affecting multiple body systems. It includes the orofacial system, which is related to vital functions such as swallowing and chewing. These functions are essential for the feeding process. **Purpose**: To identify the alterations in feeding and orofacial functions of swallowing and chewing in the child-adolescent population with Down syndrome that are described in the literature. **Methods**: In this exploratory review, advanced searches were performed in PubMed, CINAHL Complete (EBSCO), and Embase, using terms and connectors such as (down syndrome) AND (feeding disorders) NOT (review); (down syndrome) AND ((deglutition disorders) OR (dysphagia)) NOT (review); and (down syndrome) AND ((chewing disorders) OR (masticatory dysfunction)) NOT (review), among others. In total, 279 titles were identified and, after the selection process, 8 were included in the sample. After reviewing the references of the selected articles, two more studies were added. **Results**: This confirms what has been described in previous studies, according to which children with Down syndrome present alterations in feeding and in swallowing and chewing functions from an early age. **Conclusions**: It is essential that professionals detect these alterations in a timely manner. This will allow adequate treatment to be offered to patients and their

families. In this way, their quality of life can be improved. In addition, the need for an interdisciplinary approach to its management is highlighted.
Keywords: chewing; children; dentistry; Down syndrome; feeding; speech therapy; swallowing

RESUMEN

Antecedentes: El síndrome de Down es el trastorno genético más prevalente asociado a discapacidad intelectual. Se caracteriza por afectar múltiples sistemas corporales. Incluye el sistema orofacial, relacionado con funciones vitales como la deglución y la masticación. Estas funciones son esenciales para el proceso de alimentación. **Objetivo**: Identificar las alteraciones en la alimentación y las funciones orofaciales de deglución y masticación en la población infantil-juvenil con síndrome de Down que están descritas en la literatura. **Métodos**: En esta revision exploratoria se realizaron búsquedas avanzadas en PubMed, CINAHL Complete (EBSCO), y Embase, empleando términos y conectores como (down syndrome) AND (feeding disorders) NOT (review); (down syndrome) AND ((deglutition disorders) OR (dysphagia)) NOT (review); y (down syndrome) AND ((chewing disorders) OR (masticatory dysfunction)) NOT (review), entre otros. En total, se identificaron 279 títulos y, tras el proceso de selección, se incluyeron 8 en la muestra. Después, al revisar las referencias de los artículos seleccionados, se añadieron dos estudios más. **Resultados**: Se confirma lo descrito en estudios previos según los cuales los niños y niñas con síndrome de Down presentan alteraciones en la alimentación y en las funciones de deglución y masticación desde edades tempranas. **Conclusiones**: Es fundamental que los profesionales detecten estas alteraciones de manera oportuna. Esto permitirá ofrecer un tratamiento adecuado a los pacientes y sus familias. Así, se podrá mejorar su calidad de vida. Además, se destaca la necesidad de un abordaje interdisciplinario en su manejo.
Palabras Clave: alimentación; deglución; fonoaudiología; masticación; niños; odontología; síndrome de Down; terapia del lenguaje

RESUMO

Antecedentes: A síndrome de Down é a doença genética mais prevalente associada à deficiência intelectual. É caracterizada por afetar vários sistemas do corpo. Inclui o sistema orofacial, relacionado às funções vitais como deglutição e mastigação. Essas funções são essenciais para o processo de alimentação. **Objetivo**: Identificar as alterações na alimentação e nas funções orofaciais de deglutição e mastigação na população criança-adolescente com síndrome de Down descritas na literatura. **Métodos**: Nesta revisão exploratória, foram realizadas buscas avançadas no PubMed, CINAHL Complete (EBSCO) e Embase, utilizando termos e conectores como (síndrome de Down) AND (distúrbios alimentares) NOT (revisão); (síndrome de Down) AND ((distúrbios de deglutição) OR (disfagia)) NOT (revisão); e (síndrome de Down) AND ((distúrbios mastigatórios) OR (disfunção mastigatória)) NOT (revisão), entre outros. No total foram identificados 279 títulos e, após o processo de seleção, 8 foram incluídos na amostra. Posteriormente, ao revisar as referências dos artigos selecionados, foram acrescentados mais dois estudos. **Resultados**: Confirma-se o que foi descrito em estudos anteriores segundo os quais meninos e meninas com síndrome de Down apresentam alterações nas funções de alimentação e deglutição e mastigação desde tenra idade. **Conclusões**: É fundamental que os profissionais detectem estas alterações em tempo hábil. Isso permitirá que o tratamento adequado seja oferecido aos pacientes e seus familiares. Assim, sua qualidade de vida pode ser melhorada. Além disso, destaca-se a necessidade de uma abordagem interdisciplinar na sua gestão.
Palavras-chave: alimentação; crianças; engolir; fonoaudiologia; mastigar; odontologia; síndrome de Down

INTRODUCTION

Down syndrome, or trisomy 21, was first described in 1866 by Langdon Down (1). It is the most prevalent genetic disorder associated with intellectual disability. In the United States, in 2010, a prevalence of 6.7 per 10,000 live births was reported (2). In Latin America, according to the Latin American Collaborative Study of Congenital Malformations (ECLAMC), this rate is 1.88 per thousand live births (3).

This condition can affect multiple body systems, such as musculoskeletal, neurological, and cardiovascular. It is also associated with growth disorders, muscle hypotonia, atlantoaxial instability, reduced neuronal density, and cerebellar hypoplasia, among others (1). In the oral area, dentomaxillary anomalies (DMA) are the most common in people with Down syndrome (4-5). These include mandibular protrusion, anterior open bite and posterior crossbite. These are related to insufficient facial bone development and hypotonia of the orofacial and lingual muscles (6-7). Functionally, these conditions can cause orofacial dysfunctions that affect breathing, swallowing, mastication, and speech. The

alterations include oral breathing, atypical swallowing, muscle hypotonia, rhinorrhea, sialorrhea, snoring, dry lips, and lip incompetence (8).

Functional adaptations of the orofacial structure have been reported to generate a specific pattern of dentoskeletal growth and development. These adaptations seek a better integration of the musculoskeletal system and an efficient use of functions such as breathing, sucking, chewing, swallowing, speaking, and facial expressions (9-10). In children with Down syndrome, these functional adaptations, conditioned by their maxillofacial anatomy, alter the growth and development process. This causes, for example, severe changes in occlusion, favored by poor management of orofacial functions, such as swallowing and chewing, both essential for the feeding process.

Based on the background presented, the research question was formulated: What are the feeding and orofacial swallowing and chewing disorders in children with Down syndrome? To answer this question, an exploratory review of the literature was conducted with the aim of identifying the alterations in feeding and orofacial swallowing and chewing functions in the child and adolescent population with Down syndrome.

MATERIALS AND METHODS

Search Strategy and Databases

An electronic search was conducted in June 2023 using the PubMed, CINAHL Complete EBSCO, and Embase databases. The search terms used are detailed in Table 1.

TABLE 1
Search Terms and Boolean Connectors Used in Three Databases

Database	PubMed	CINHAL Complete EBSCO	EMBASE
Search Strategy 1	(Down Syndrome AND Deglutition disorders OR Dysphagia) NOT (review)	Down syndrome AND Deglutition disorders OR Down Syndrome AND Dysphagia NOT review	(Down Syndrome AND Deglutition disorders OR Dysphagia) NOT (review)
Search Strategy 2	(Down Syndrome and feeding disorders) NOT (review)	Down Syndrome and feeding disorders NOT review	(Down Syndrome and feeding disorders) NOT (review)
Search Strategy 3	(down syndrome) AND ((chewing disorders) OR (masticatory dysfunction)) NOT (review)	Down syndrome AND chewing disorders OR down syndrome AND masticatory dysfunction NOT review	(Down syndrome) AND ((chewing disorders) OR (masticatory dysfunction)) NOT (review)

Source: the authors.

Eligibility Criteria

As an inclusion criterion, articles were selected that were primary sources, published in English or Spanish, and that addressed swallowing and chewing disorders in children with Down syndrome, that is, in individuals under 18 years of age. To analyze recent literature, articles published between 2013 and 2023 were included. Reviews were excluded.

Register of Studies and Synthesis of Results

After conducting the search in line with the previously established inclusion and exclusion criteria, the titles and abstracts of all identified articles were reviewed. Articles unrelated to the research question

and duplicates were excluded. The full text of the selected articles was then analyzed. The objectives, the evaluated population, the methodology used, and the outcomes measured were described. Studies deemed irrelevant were removed. These included studies on adult populations with Down syndrome or other conditions. It also included investigations validating instruments for assessing swallowing and chewing alterations without specifically addressing disorders of these functions. This process resulted in a definitive list. A consolidated dataset was developed, classifying the articles by topic. This allowed addressing each aspect considered in the research question.

RESULTS

Selection of Studies

The initial electronic search yielded 279 results. After removing 8 duplicates, 271 articles remained for evaluation in the selection stage. Following the analysis of titles and abstracts, 257 studies were excluded for not being related to the topic of this review. Thus, 14 articles were selected for full-text reading. Finally, 8 articles were included in the review (Table 2), as they were directly related to the research question.

Additionally, the references of the selected articles were analyzed. Based on their titles, 2 studies not identified in the initial literature search were found. These are presented in Table 3. A full-text reading of both articles was conducted for their inclusion. Since they were related to the topic under study, they were added to the final list. Figure 1 shows the process of study identification and selection using a flow diagram. Table 2 presents the 8 selected studies along with their main characteristics.

The 8 selected studies were conducted in 4 countries: India by Anil, *et al.* (14); the United States by Jackson, *et al.* (15), Cochran, *et al.* (17), and Simons, *et al.* (18); Brazil by Sales, *et al.* (19), Pinheiro, *et al.* (21), and Bermúdez, *et al.* (15); and the Netherlands by In't Veld, *et al.* (20).

The types of studies identified and selected included the following methodologies, as reported by the authors: questionnaire design and validation by Anil, *et al.* (14); retrospective, observational, descriptive, and cross-sectional study by Bermúdez, *et al.* (15); retrospective and observational studies by Jackson, *et al.* (15) and Simons, *et al.* (18); qualitative study by Cochran, *et al.* (17); case series by Sales, *et al.* (19); descriptive study by In't Veld, *et al.* (20); and quantitative, longitudinal, and interventional study by Pinheiro, *et al.* (21).

The studies selected for this review were published between 2016 and 2022. All the research focused on the clinical and functional characteristics of swallowing and chewing practices in children with Down syndrome.

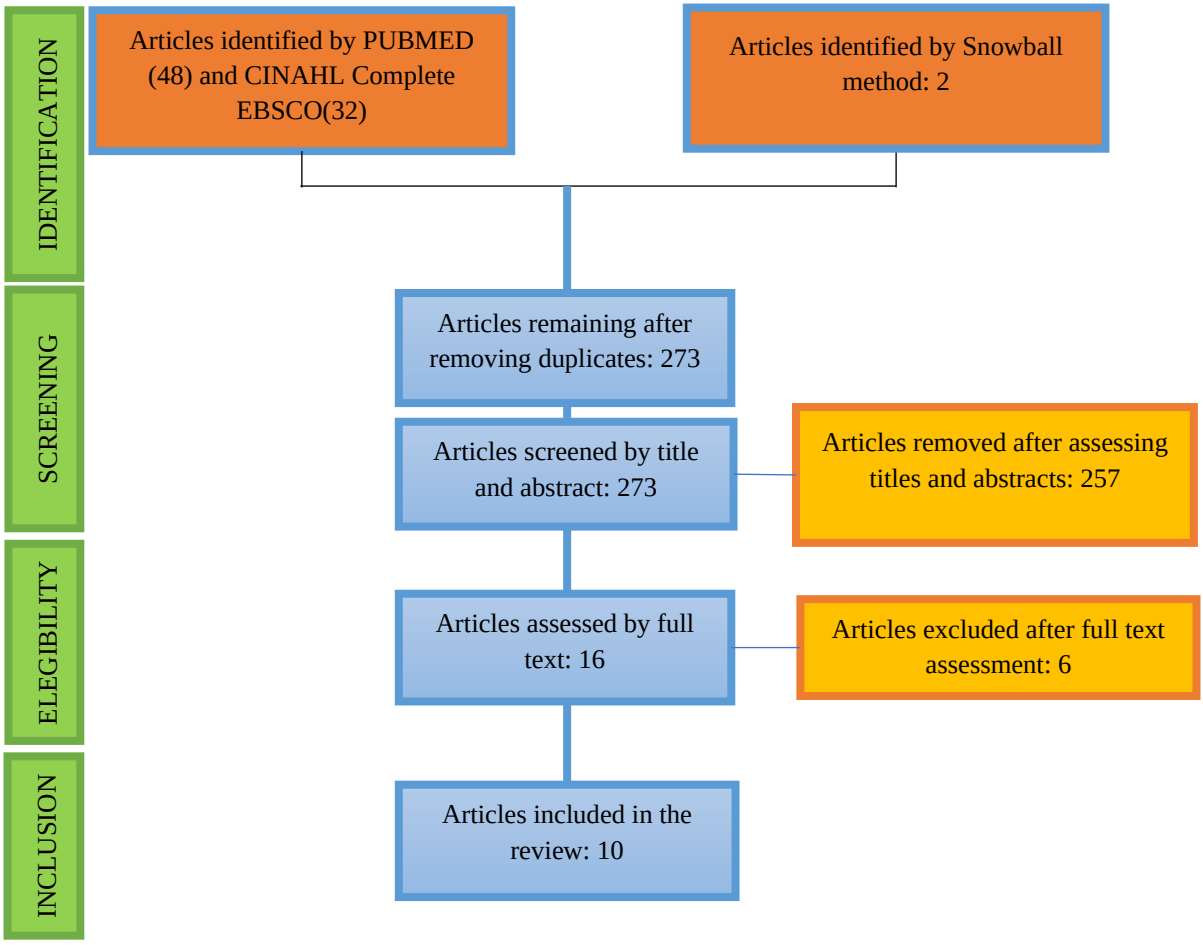

FIGURE 1
Diagram of the Process for Identification and Selection of Titles for the Review

TABLE 2
Characteristics of the Studies Selected for Review

Author(s), Year	Population, Country	Methods	Outcome
Anil, *et al*., 2019 (14)	Parents of children with Down Syndrome, between 2 and 7 years old, India.	Validated questionnaire application.	Feeding and swallowing problems present in children between 2 and 7 years old.
Bermudez, *et al*., 2019 (15)	People with Down Syndrome. 0 – 5 years old: 187 children, 6 – 10 years old: 442 children, 11 – 20 years old: 427 children. / Down Syndrome Outpatient Clinic, Hospital de Clínicas, Universidade Federal do Paraná, Curitiba, Paraná, Brazil.	Retrospective observational, descriptive and cross – sectional study.	Gastrointestinal disorders prevalence in people with Down Syndrome in a 10 year – period.
Jackson, *et al*., 2019 (16)	Children with Down Syndrome up to 12 months old. / Anna and John J. Sie Center for Down Syndrome, Children Hospital, Denver, Colorado, USA.	Retrospective observational study.	Association amongst age, dysphagia and medical comorbidities in infants with Down Syndrome.
Cochran, *et al*., 2022 (17)	Parents of children with Down Syndrome, EE.UU.	Qualitative study	Family experiences associated with complementary feeding.
Simons, *et al*., 2016 (18)	Children with laryngomalacia, who attend the Aerodigestive Center, University of Pittsburgh, USA.	Retrospective study	Dysphagia prevalence in children with laryngomalacia and its relationship with swallowing disorders.
Sales, *et al*., 2017 (19)	Children with Down syndrome between 4 and 17 months old, Brazil.	Report of a series of clinical cases.	Qualitative and quantitative analysis of oropharyngeal swallowing in children with Down Syndrome.
In't Veld, *et al*., 2020 (20)	Children with Down syndrome, Donders Centre for Neuroscience, Department of Rehabilitation, Radbound University Medical Center, Nijmegen, Netherlands.	Descriptive study and application of two tests to evaluate chewing efficiency and resistance.	Chewing problems associated with efficiency and resistance.
Pinheiro, *et al*., 2018 (21)	Children and young people with Down syndrome, Speech and Language Department, Health Sciences Center, Universidade Federal da Paraíba, Joao Pessoa, Brazil.	Quantitative, longitudinal, and interventional study.	Electrostimulation effects on orofacial muscles and on chewing, breathing and swallowing in people with Down Syndrome.

TABLE 3
Articles Selected Using the Snowball Technique

Author(s), Year	Population, Country	Methods	Outcome
Jackson, *et al*. 2016 (22)	158 children with Down Syndrome, Anna and John J. Sie Center for Down Syndrome, Children´s Hospital, Denver, Colorado, EE.UU.	Retrospective study	Identification of the characteristics of dysphagia in the oral and pharyngeal phase of children with Down syndrome.
O'Neill & Richter, 2013 (23)	212 children with Down Syndrome, Arkansas Children's Hospital, University of Arkansas for Medical Sciences, EE.UU.	Retrospective study	Evaluation of the magnitude of the incidence of pharyngeal dysphagia in a cohort of children with Down syndrome.

DISCUSSION

After analyzing the articles selected for this review, three main aspects were identified for study: feeding disorders in children with Down syndrome, swallowing disorders, and masticatory function disorders.

Regarding feeding disorders, Anil, *et al.* (14) analyzed feeding and swallowing problems in children with Down syndrome, assessing their physical, functional, and emotional impact. The findings indicated that children with trisomy 21 experienced greater feeding difficulties compared to those with neurotypical development. These difficulties were attributed to oral hyposensitivity, manifested as altered oral proprioception, with an immature chewing pattern being the primary issue during the oral phase. Functional complications were also observed, such as food leakage through the lips, aversion to specific foods (particularly those with challenging textures), the need for a specific feeding position, and the requirement to push food to the back of the mouth, among others. On an emotional level, frustration was reported due to dependency on others during social situations, food rejection, and undesirable behaviors during feeding.

In a qualitative study, Cochran, *et al.* (17) observed that caregivers and families required support for the introduction of complementary feeding. It was reported that children experienced difficulties at three key stages: initiating breastfeeding or bottle-feeding, introducing complementary foods, and incorporating foods requiring chewing. Parents reported feeling a high level of stress when facing these stages without adequate information provided by healthcare personnel.

Regarding swallowing disorders, Anil, *et al.* (14) reported that children with Down syndrome experienced difficulties due to the lack of anterior seal and poor sweeping action of the tongue, which impaired efficient bolus transit. The most frequent issues during the oral phase included the absence of mandibular closure after food intake, difficulty performing lateral tongue movements (leading to food accumulation in the lateral and anterior grooves), and reduced anteroposterior tongue peristalsis with uncoordinated movements, which hindered bolus formation and control. Additionally, a high number of oral-motor problems were observed compared to the neurotypical population. This could be attributed to hypotonia of the oral structures, resulting in an open-mouth posture, poor mandibular closure, lack of firm lip seal for suction, difficulty in gradual jaw mobilization, and tongue and lip movements affecting swallowing.

Another study evaluated video fluoroscopy reports to describe the characteristics of dysphagia during the oral phase in children with Down syndrome. The findings indicated that this population experiences oral motor dysfunction characterized by weak lip closure, a compression pattern without generating intraoral negative pressure, tongue protrusion, and chewing difficulties. Additionally, a lack of coordination in the breathing-sucking-swallowing triad was reported. Less frequent features included fatigue, slowed oral transit, and post-swallow oral residue. Oral sensory difficulties were also observed, with oral hypersensitivity being particularly notable (22).

In a study conducted by Sales, *et al.* (19), which analyzed a case series of children with Down syndrome aged 4 to 17 months, all participants were found to have oral incoordination and oral residue during the oral phase of swallowing. Additionally, five participants exhibited labial sphincter incompetence.

Regarding the pharyngeal phase, Anil, *et al.* (14) reported that the most frequent issues in the studied population included food retention in the mouth after chewing without swallowing, which could be attributed to a delay in initiating the oropharyngeal swallowing movement. Additionally, cases of aspiration and/or choking during liquid intake were identified. Some participants also reported food sticking in the pharynx or described a burning sensation in the mouth and throat.

In their study, O'Neill and Richter (23) evaluated the incidence of pharyngeal dysphagia in a large population of children with Down syndrome. They found that approximately 60 % had pharyngeal dysphagia confirmed by video fluoroscopy. This highlights that swallowing disorders during the pharyngeal phase are common and require further study to identify risks and define management

strategies. They also noted that these patients often have medical comorbidities, aerodigestive disorders, and neurological conditions that affect swallowing function, significantly influencing pharyngeal dysphagia.

In another study, Jackson, *et al.* (22) described the characteristics of pharyngeal dysphagia in children with trisomy 21 through video fluoroscopic analysis. The researchers identified that the main complications included aspiration (primarily silent), deep laryngeal penetration, pharyngeal residue, nasopharyngeal regurgitation, and, in a small percentage, delayed glottic trigger.

A group of researchers evaluated the associations between age, dysphagia, and medical comorbidities in young children with Down syndrome. In the studied sample, they found that nearly one-third of children under six months and more than 50 % of children aged six to twelve months exhibited deep laryngeal penetration and/or aspiration. Additionally, cases of nasopharyngeal regurgitation and pharyngeal residue were reported. They identified conditions associated with dysphagia, such as laryngomalacia, pulmonary hypertension, pneumonia, and congenital heart disease (16). However, in a previous study published by Simons, *et al.* (18), it was concluded that there is no significant relationship between the presence of Down syndrome and the severity of laryngomalacia or swallowing dysfunction. On the other hand, in a case series by Sales, *et al.* (19), pharyngeal phase alterations, such as posterior oral spillage and laryngotracheal penetration and/or aspiration, were identified.

Among the commonly observed problems during the esophageal phase is vomiting after eating. This could be attributed to swallowing partially chewed or unchewed bites, as well as reduced muscle tone, which promotes poor digestion, esophageal obstruction, or gastroesophageal reflux (14–15).

On the other hand, Bermúdez, *et al.* (15) analyzed the medical records of 1,207 patients with Down syndrome over 10 years and reported a significantly lower incidence of dysphagia compared to other studies. The authors attributed these findings to the fact that the subjects received stimulation from birth, and 75.2 % of the children were successfully breastfed, with an average breastfeeding duration ranging from 6 to 48 months. They also observed that parents, guided by a healthcare team, introduced solid foods around 6 months of age, promoting normal oral motor development. As a result of these practices, fewer anatomical changes and reduced oral hypersensitivity were evident, conditions that are typically common in this syndrome and can lead to chewing impairments and difficulty accepting solid foods with a pasty consistency.

Regarding masticatory function, In't Veld, *et al.* (20) reported that, in terms of efficiency, children with Down syndrome required more chewing cycles and performed more swallowing acts to manage a cookie compared to neurotypical children of the same age. However, no differences were found in masticatory endurance between the two groups. For these findings, they analyzed the feasibility of applying two tests: TOMASS and 6MMT, designed to evaluate chewing in children with Down syndrome. The TOMASS (Test of Mastication and Swallowing of Solids) provides information about masticatory efficiency (24), while the 6MMT (6-Minute Mastication Test) assesses masticatory endurance by comparing observations at 1 minute and 6 minutes (25). The authors concluded that both tests are applicable to the pediatric population with trisomy 21, though TOMASS is more significant for evaluating chewing problems and measuring the effectiveness of exercises prescribed to address these issues.

On the other hand, Anil, *et al.* (14) described that the most prevalent problem during the oral phase of swallowing was an immature chewing pattern. They also observed that food remained in the mouth longer without being chewed. Additionally, they reported that children with Down syndrome took longer to manipulate the bolus, demonstrating inadequate oral transit time.

Finally, Pinheiro, *et al.* (21) investigated the effects of functional electrical stimulation (FES) on orofacial musculature and the functions of chewing, breathing, and swallowing. Regarding chewing, they observed that the combination of FES with masticatory therapy enabled the achievement of lip seal without perioral or facial muscle tension. Additionally, they achieved alternating and bilateral molar chewing patterns, as well as the use of incisors to cut food, showing a tendency toward the standards observed in the neurotypical population.

CONCLUSIONS

Down syndrome is associated with widely described physical and cognitive characteristics. However, there is limited research focusing on the study of routines and processes related to feeding.

Regarding swallowing, children with Down syndrome primarily experience difficulties during the oral phase and, to a lesser extent, during the pharyngeal and esophageal phases. For this reason, it is essential to screen all children with this condition to detect orofacial function disorders early.

Regarding chewing, children with Down syndrome face difficulties in performing the different stages of this process. Therefore, they require support from the start of complementary feeding to develop the skills needed to properly manage foods with challenging textures.

According to the analyzed reports, providing early therapy could improve the treatment prognosis for the difficulties observed in feeding practices as well as in swallowing and chewing functions.

RECOMMENDATIONS

An analysis of the published literature identified aspects that have not been thoroughly studied. Among these is the need to create clinical guidelines to standardize the approach to disorders associated with the feeding process. These guidelines should include strategies to support parents through the different developmental stages, promoting the introduction of foods with various textures without delaying the consumption of solid and diverse foods. Additionally, the importance of addressing feeding not only from a nutritional perspective but also as an opportunity to encourage maxillofacial growth and development is emphasized.

It is recommended to investigate how family habits, as well as the health perceptions and beliefs of primary caregivers, influence the feeding process of children with Down syndrome. This will enable healthcare professionals to implement interventions with a biopsychosocial approach, considering the environment in which the child is raised. The goal is to promote the generalization of expected behaviors. Additionally, caregivers should have access to updated, evidence-based information to ensure timely therapies tailored to the reality of each patient and their family.

In this same vein, it is suggested to develop clinical guidelines or protocols to be implemented immediately after the birth of a child with Down syndrome. The goal is to initiate early therapy, considering the significant benefits of receiving timely rehabilitation.

Regarding feeding, swallowing, and chewing disorders, it is essential to establish interdisciplinary teams that promote the generation of new knowledge through collaborative research. This approach will enable the design of comprehensive interventions that address all the needs of children with Down syndrome.

From a biopsychosocial perspective, there are few reports addressing the activity and participation difficulties stemming from feeding disorders, as the biomedical approach predominates. Therefore, it is important to analyze how feeding can positively or negatively impact the development of individuals with trisomy 21, considering that it is a fundamental daily activity and plays a key role in social interaction.

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