






Dysphagia in Autism Spectrum Disorder (ASD): An integrative review

Disfagia no Transtorno do Espectro Autista (TEA): uma revisão integrativa

Disfagia en el trastorno del espectro autista (TEA): una revisión integradora

Brenda Carla Lima Araujo¹ 
Keila Suyane de Jesus Felix¹ 
Kawanny Oliveira Marques da Silva¹ 
Rosana Carla do Nascimento Givigi¹ 
Susana de Carvalho¹ 

Abstract

Introduction: Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social communication, changes in sensitivity and eating difficulties. **Objective:** To carry out an integrative review of swallowing changes in individuals with ASD. **Methods:** The research was carried out through a search for national and international articles, using descriptors for the research, as well as inclusion and exclusion criteria for selecting the final sample. The PPOT strategy was used to define eligibility criteria, including population (children and adults), predictor (ASD diagnosis), outcome (report or diagnosis of dysphagia oral, pharyngeal or esophageal), and study type (observatory studies). The search was carried out from June to August 2023, in the databases: Pubmed, Scopus, Embase and Google Scholar. **Results:** Ten studies were selected with patients diagnosed with ASD who reported symptoms of oropharyngeal and esophageal dysphagia, in addition to complaints about food intake. Studies suggest that children with ASD may present problems with oral motor dysfunction, inadequate eating frequency, obsessive eating patterns, specific presentation of certain foods, food selectivity and sensory processing difficulties. **Conclusion:** It is concluded that there is no robust scientific evidence about the presence of dysphagia in patients with ASD.

Keywords: Autism Spectrum Disorder; Deglutition; Deglutition disorder; Dysphagia.

¹ Universidade Federal de Sergipe, SE, Brazil.

Authors' contributions:

BCLA and KSJF: participated in the conception of the study, collection, analysis and interpretation of data and writing of the article. KWOMS, RCNG and SC: participated in the final writing of the article.

E-mail address: Brenda Carla Lima Araujo - brendaaraujo@yahoo.com.br

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Resumo

Introdução: O Transtorno do Espectro Autista (TEA) é um distúrbio do neurodesenvolvimento caracterizado por déficits na comunicação social, alterações de sensibilidade e dificuldades alimentares. **Objetivo:** Realizar uma revisão integrativa das alterações de deglutição em indivíduos com TEA. **Métodos:** A pesquisa foi realizada por meio de uma busca por artigos nacionais e internacionais, utilizando descritores para a pesquisa, bem como critérios de inclusão e exclusão para a seleção da amostra final. A estratégia PPOT foi utilizada para definir critérios de elegibilidade, incluindo população (crianças e adultos), preditor (diagnóstico de TEA), desfecho (relato ou diagnóstico de disfagia oral, faríngea ou esofágica) e tipo de estudo (estudos observatórios). A busca foi realizada no período de junho a agosto de 2023, nas bases de dados: Pubmed, Scopus, Embase e Google Scholar. **Resultados:** Foram selecionados dez estudos com pacientes diagnosticados com TEA que relataram sintomas de disfagia orofaríngea e esofágica, além de queixas sobre ingestão alimentar. Os estudos sugerem que crianças com TEA podem apresentar algum problema de disfunções motoras orais, frequência alimentar inadequada, padrões alimentares obsessivos, apresentação específica de determinados alimentos, seletividade alimentar e dificuldades de processamento sensorial. **Conclusão:** Conclui-se que não há evidências científicas robustas sobre a presença de disfagia em pacientes com TEA.

Palavras-chave: Transtorno do Espectro Autista; Deglutição; Transtorno de deglutição; Disfagia

Resumen

Introducción: El Trastorno del Espectro Autista (TEA) es un trastorno del neurodesarrollo caracterizado por déficits en la comunicación social, cambios en la sensibilidad y dificultades alimentarias. **Objetivo:** Realizar una revisión integradora de los cambios en la deglución en individuos con TEA. **Métodos:** La investigación se realizó mediante una búsqueda de artículos nacionales e internacionales, utilizando descriptores para la investigación, así como criterios de inclusión y exclusión para la selección de la muestra final. La estrategia PPOT se utilizó para definir los criterios de elegibilidad, incluida la población (niños y adultos), el predictor (diagnóstico de TEA), el resultado (informe o diagnóstico de enfermedad oral, faríngea o esofágica) y el tipo de estudio (estudios observatorios). La búsqueda se realizó de junio a agosto de 2023, en las bases de datos: Pubmed, Scopus, Embase y Google Scholar. **Resultados:** Se seleccionaron diez estudios con pacientes diagnosticados de TEA que refirieron síntomas de disfagia orofaríngea y esofágica, además de quejas sobre la ingesta de alimentos. Los estudios sugieren que los niños con TEA pueden presentar problemas de disfunción motora oral, frecuencia inadecuada de alimentación, patrones alimentarios obsesivos, presentación específica de ciertos alimentos, selectividad alimentaria y dificultades en el procesamiento sensorial. **Conclusión:** Se concluye que no existe evidencia científica robusta sobre la presencia de disfagia en pacientes con TEA.

Palabras clave: Trastorno del Espectro Autista; Deglución; Trastorno de la deglución; Disfagia

Introduction

Autism Spectrum Disorder (ASD) is defined by changes in social interaction skills, as well as difficulties in communication and a tendency to repetitive and stereotypical behaviors¹. This disorder has its origins in childhood and often lasts throughout adolescence and adulthood, with most diagnoses occurring in the first five years of life².

Feeding difficulties in children appear to be increasingly on the rise over time, possibly due to the increased survival rate of high-risk premature

newborns with developmental delays, as well as children diagnosed with ASD³. Studies suggest that, in children without impairments, around 25% to 45% difficulties in the eating process. On the other hand, for children with developmental impairment, this rate varies between 33% and 80%^{3,4}. Eating difficulties can be linked to different clinical causes such as neurological disorders, congenital diseases, oral, nasopharyngeal and esophageal motor dysfunction, as well as food allergies and gastrointestinal disorders, proving to be a common disorder in many individuals with ASD⁵.



During the feeding process, several structures play crucial roles, including the oral cavity, pharynx, larynx, and esophagus. Changes that affect these structures can result in dysphagia, defined as any difficulty in transporting food and saliva in an intact manner, ranging from the oral preparatory phase, involving chewing and efficient positioning of the food bolus, to the transport phase, pharyngeal and esophageal^{6,7}.

Eating issues in ASD are described in the literature, with the presence of changes characterized by high restriction in eating, including problems such as food selectivity, oral motor dysfunction, compulsive eating patterns, inadequate food quantity, food texture and aroma in a standardized way, with dysphagia being a difficulty also common in individuals with ASD⁶. A systematic review with meta-analysis found an association between the physiology of ASD and eating and behavioral difficulties. These findings suggest a direct or indirect relationship with problems with food intake and may include impairment in sensory processing, changes in oral motor skills such as chewing and swallowing or digestive disorders⁸.

For these reasons, it is important to understand the difficulties in feeding this population, including the swallowing function, in addition to the impact on the health, treatment and quality of life of people with ASD. Therefore, the objective of the present study was to carry out an integrative review of the literature on changes in swallowing in ASD.

Method

The methodology of this study was characterized as an integrative review of the scientific literature. This method allows, through studies already carried out, to determine current knowledge on a specific topic, as it is aimed at identifying, analyzing and synthesizing results of independent studies on the same topic⁹. Initially, the guiding question of this study was: “What scientific evidence is available on dysphagia in people with ASD?”

The PPOT strategy (population, predictor, outcome and type of study) was used to define the

eligibility criteria: (P) population: children and/or adults, regardless of age and sex; (P) predictor: diagnosis of ASD (O) outcome: report and/or diagnosis of oral, pharyngeal or esophageal dysphagia and (T) type of study: observational studies.

The search was carried out from June to August 2023, being limited to studies published in full version, without language and date restrictions. The databases consulted were: Pubmed, Scopus, Embase and Google Scholar. The structured search strategy was directed to each database used, with the aim of covering the largest possible number of studies, using the following descriptors and their variations: Autism Spectrum Disorder; Autistic Spectrum Disorder; Swallowing Disorders; Swallowing Disorders; Oropharyngeal Dysphagia; Dysphagia, Oropharyngeal; Esophageal Dysphagia; Esophageal; Swallowing. All terms used are registered in the Health Sciences Descriptors (DECs) and Medical Subject Headings (MeSH). For articles not available in electronic databases or for data not available in the articles included in this review, the authors were contacted to obtain the necessary information. The first 100 results were analyzed on Google Scholar. (Table 1).

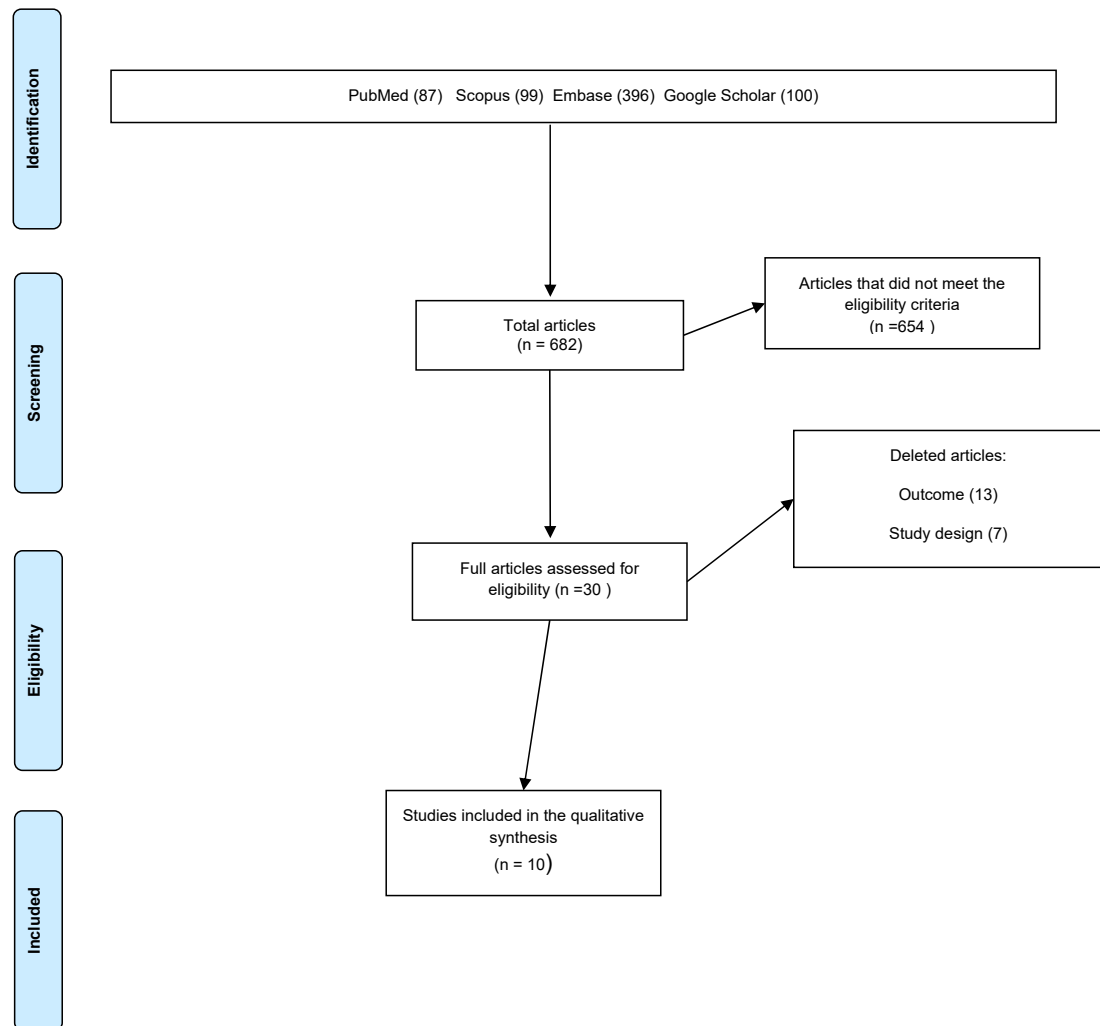
In this review, editorials, comments and opinions, reflection articles, projects and technical reports, review articles and articles that dealt with other changes that were not related to the topic of ASD and dysphagia were excluded. To expand the number of eligible articles retrieved, no filters were used in the search. Observational cross-sectional, case-control and cohort studies were included.

After the final selection of the researched studies included in the analysis, the main information was compiled. A descriptive analysis was carried out and a standard form was used containing information: authors / year / country, type of study, objective and results.

In the initial search, 684 articles were found. With the selection by title and abstract, 30 studies were considered potentially relevant to be read in full. Finally, 10 articles met the eligibility criteria and were included in this integrative review (Figure 1).

Table 1. Database search strategy

Data base	Search strategies	Nº
Embase	"Autism Spectrum Disorder"; "Autism Spectrum Disorders"; "Autistic Spectrum Disorder"; "Autistic Spectrum Disorders"; " Disorder, Autistic Spectrum AND Deglutition"; "Deglutition Disorders"; " Swallowing"; " Dysphagia Oropharyngeal"; "DysphagiaEsophageal"; " Deglutition".	396
Scopus	"Autism Spectrum Disorder "; "Autistic Spectrum Disorder" AND "Deglutition Disorders"; "Swallowing Disorder"; "Dysphagia"; "Esophageal Dysphagia"; "Deglutition"; "Swallowing".	99
Pubmed	Autism Spectrum Disorder[Mesh] AND Deglutition Disorders[Mesh]; "Swallowing Disorders"; "Dysphagia Oropharyngeal"; " Esophageal Dysphagia"; " Deglutition[Mesh]".	87
Google Scholar	"Autism Spectrum Disorder"; "Spectrum AND Deglutition Disorders"; "Deglutition Disorders"; "Swallowing Disorder"; "Dysphagia Oropharyngeal"; "Esophageal Dysphagia".	100
TOTAL		682

**Figure 1.** Flowchart of the article search process

Results

The ten studies selected in this integrative review, the United States of America was the country with the highest number of publications with five studies¹⁰⁻¹⁴, followed by the other countries: Sweden, Turkey, Brazil, Egypt and South Africa, which totaled five publications¹⁵⁻¹⁹. The articles are summarized with a description of the authors, year of publication, country, type of study, objective and results (Chart 1).

The articles were published between the years 2014 and 2022. Of these studies, five are cohort studies^{12,16,14,18,19}, two clinical cases^{10,13}, one longitudinal descriptive¹⁷, one case-control¹¹ and one cross-sectional study¹⁵. The classification of each study was approached according to the description of its own authors.

The sample of studies included in this review were individuals evaluated and clinically diagnosed with ASD. The results demonstrated that three articles informed the protocols used to obtain the diagnosis: the Autism Diagnostic Ob-

servations Scale (ADOS-2), American Psychiatric Association 2013 (DSM-5) and Autism Diagnostic Interview-Revised (ADI-R) were the instruments cited for ASD screening applied by neurologists, psychiatrists and/or multidisciplinary teams^{11,17,14}.

Among the findings, only the study carried out by Kamionkowski et al. (2021)¹² approached adult patients, the other articles preferred to approach the child population, distributed between ages 28 months and 12 years.

Oral feeding is used by the majority of patients with ASD, it was found together with feeding problems, including changes in food preparation, chewing and swallowing. Regarding the use of a nasogastric tube in pediatric patients, only one article addressed the use of a mixed feeding route, that is, combined oral feeding and nutritional supplementation through a tube¹³.

Patients reporting symptoms of oropharyngeal and esophageal dysphagia were the focus of most studies. The most common complaints were swallowing and ingestion problems^{15,11,13,17,14,18,19}. A summary of the studies included in this review is presented in Chart 2.

Chart 1. Bibliographic data of the articles selected in the review (n=10).

Year	Publication	Title	Authors
2014	J. Autism Dev Disord.	Gastrointestinal problems in children with autism, developmental delays or typical development.	Chaidez V.; Hansen R. L.; Hertz-Picciotto I.
2014	Behav Modif.	Multi-component treatment to reduce packing in children with feeding and autism spectrum disorders	Levin D. S.; Volkert V. M.; Piazza C. C. A.
2016	Case Rep Psychiatry.	Targeted Nutritional and Behavioral Feeding Intervention for a Child with Autism Spectrum Disorder	Barnhill K; Tami, A; Schutte, C; Hewitson, L; Olive, M.L.
2019	Frontiers in pediatrics	Feeding Problems Including Avoidant Restrictive Food Intake Disorder in Young Children With Autism Spectrum Disorder in a Multiethnic Population.	Nygren, G.; Linnsand, P.; Hermansson, J.; Dinkler, L.; Johansson, M.; Gillberg, C.
2020	Autism Res.	Developmental-behavioral profiles in children with autism spectrum disorder and co-occurring gastrointestinal symptoms.	Restrepo B.; Angkustsiri K.; Taylor S.L.; Rogers S.J; Cabral J; Heath B; Hechtman A; Solomon M; Ashwood P; Amaral D.G; Nordahl C.W.
2020	African Health Sciences.	Parent-reported feeding and swallowing difficulties of children with Autism Spectrum Disorders (aged 3 to 5 years) compared to typically developing peers: a South African study.	Viviers, M.; Jongh, M.; Dickonson, L.; Malan, R.; Pike, T.
2021	Dysphagia	A comparative analysis of chewing function and feeding behaviors in children with autism	Şahan, A.K.; Öztürk, N.; Demir, N.; Karaduman, A. A.; Serel Arslan, S.
2021	Neurogastroenterology & Motility.	The relationship between gastroesophageal reflux disease and autism spectrum disorder in adult patients in the United States.	Kamionkowski S; Shibli, F; Ganocy, S; Fass, R.L.
2021	Physis.	Food and nutritional aspects of children and adolescents with autism spectrum disorder.	Magagnin, T.; Da Silva, M. A.; De Souza Nunes, R. Z.; Ferraz, F.; Soratto
2022	Egypt J. Otolaryngol.	Screening of gastrointestinal symptoms and celiac disease in children with autism spectrum disorder	Ahmed, E; Mansour, A; Amer, A; Barakat, T; Baz.

Chart 2. Evidence found on dysphagia in ASD

Authors	Year Country	Methods	Objective	Sample size	Results
Chaidez; hansen; hertz..	2014/ United States	Case-control study	Compare and gastrointestinal problems in a group of children with ASD, developmental delay and typical development.	960 children were included (ASD n=499, developmental delay n= 324 and typical children= 137).	Children with ASD were at least three times more likely to present gastrointestinal symptoms, including: abdominal pain, pain when defecating, constipation, gas, diarrhea, sensitivity to food, difficulty swallowing and vomiting.
Levin; Volkert; Piazza.	2014/ United States	Descriptive clinical case study	Understand two cases of children with eating disorders for the behavioral treatment of packaging (holding food without swallowing in the oral cavity).	2 female children, both 4 years old.	The children presented oral motor inability to form an effective food bolus for swallowing. Efficacy was observed when combining multicomponent treatments to facilitate swallowing.
Barnhill et al.	2016/ United States	Descriptive clinical case study	To describe the results of an intensive short-term nutritional and behavioral outpatient feeding program for a child with ASD.	A 28-month-old female participant previously diagnosed with ASD.	A patient with ASD presented frequent choking during meals.
Nygren G et al.	2019/ Sweden	Longitudinal study	To examne eating problems in preschool children with ASD.	46 children (9 girls, 37 boys) born between 2010 and 2016 and diagnosed with ASD were included.	Of the 46 children with ASD, five had oral motor difficulties and 48% reported swallowing problems.
Restrepo et al.	2020/ United States	Cohort study	To determine the frequency of gastrointestinal symptoms in preschool-aged children diagnosed with ASD compared to typically developing (TD) children of the same age.	313 children, 255 children with ASD (184 men, 71 women) and 129 without ASD (75 men, 54 women).	In children with ASD, a greater number of gastrointestinal symptoms were identified, such as abdominal pain, vomiting, blood in the stool and difficulty swallowing associated with self-injurious behaviors and somatic complaints.
Viviers et al.	2020/ South Africa	Cohort study	To investigate eating and swallowing difficulties in children aged 3 to 5 years with ASD compared to children with typical development.	21 children with ASD and 21 with typical development (aged between 3 and 5 and 11 years) were included.	The BAMBI protocol, 53% of children diagnosed with ASD had severe eating and swallowing difficulties compared to neurotypical children.
Sahan et al.	2021/ Turkey	Cohort study	Compare the level of chewing performance and analyze eating behaviors in children with ASD and children with typical development.	37 children with autism and 19 classmates with typical development between the ages of 4 and 12.	Children with ASD performed worse at mealtime compared to typical children, negatively affecting swallowing and the quality of life of individuals and their families.
Kamionkowski et al.	2021/ United States	Descriptive study Cohort study	To determine and evaluate the relationship between gastroesophageal reflux disease (GERD) and autism in adults and its complications	840 were included (410 without ASD and 430 with ASD)	Patients with ASD were more likely to develop GERD, including symptoms such as coughing, pain in the throat/ swallowing, sleep apnea, hoarseness and chest pain.
Magagnin et al	2021/ Brazil	Cohort study	To evaluate the habits, difficulties and strategies when feeding children and adolescents diagnosed with ASD.	The study was carried out with 14 parents of children and adolescents with ASD.	Study observes diversified diet with dysfunctional oral motor habits that affect sensory activities in eating performance.
Ahmed et al.	2022/ Egypt	Cross-sectional observational study	To evaluate the symptomatological manifestations of gastrointestinal disorders in children with ASD.	114 children with ASD over 3 years old.	3 of the participants had difficulty swallowing as a possible symptom of gastrointestinal problems, which are more common in children with ASD.

Discussion

This literature review sought to identify swallowing changes in ASD. Most studies demonstrated the comparison of problems related to eating and swallowing difficulties between typical people and those with ASD^{11,12,14,18,19}. In research carried out by Viviers et al. (2020)¹⁹ with 42 children (21 individuals with ASD and 21 typically developing) it was demonstrated that the severity of dysphagia in individuals with ASD was 48%, while typical children presented 30% of swallowing difficulties.

Likewise, an investigation carried out by Restrepo and other researchers (2020)¹⁴ with 255 children with ASD and 129 typical children, demonstrated that 9% of children with ASD had swallowing problems, compared to reports from children without ASD, who did not present problems at the time of feeding. On the other hand, within the studies selected in this review, one study did not show a significant difference between swallowing difficulties and ASD. Chaidez, Hansen and Hertz (2014)¹¹ obtained samples from only 4.2% of 499 children with ASD, while the percentage of children without ASD (group with 324 children) was only 0.3%.

The presence of frequent choking and coughing are possible characteristics suggestive of dysphagia. Dysphagia is a disorder that affects the ability to swallow safely and effectively. It can have varied causes that directly affect the individual's life and routine, altering not only the intake of the bolus, but also the phases of swallowing³. Diagnosing dysphagia in people with ASD may involve a comprehensive assessment carried out by a multidisciplinary team, including doctors, speech therapists and other healthcare professionals. Some of the common methods used to diagnose dysphagia in individuals with ASD include history and clinical examination, clinical assessment of swallowing and objective examinations³.

A clinical case study carried out with a 28-month-old patient previously diagnosed with ASD and food selectivity, obtained information about her eating routine, where the patient presented a frequency of choking followed by emesis when eating, with foods other than breast milk. Despite these findings, the authors reinforce the lack of evidence on ASD and dysphagia and suggest oral motor assessment and swallowing function in children with ASD¹⁰.

Evidence indicates that sucking, chewing and swallowing problems can generate resistance to certain food textures, being more costly for children with ASD who have difficulty transitioning to solid foods¹⁸. In a careful analysis carried out by Sahan et al. (2021)¹⁸, children with ASD presented a percentage of 47.2% with difficulties in chewing, in the stages of crushing and pulverizing solid food, compared to children with typical development with only 10.5% in changes in the transitional stage of chewing. There was a significant difference between the groups with mean scores on the Karaduman Chewing Performance Scale (KCPS) of children with ASD and children with typical development (0.58 ± 0.60 and 0.10 ± 0.31 respectively) indicating the need for greater attention to individuals with ASD as they can negatively affect food acceptance, causing restriction and limitation of the food repertoire.

A study included in this review classifies eating difficulties as: oral motor dysfunctions, inadequate eating frequency, obsessive eating patterns, specific presentation of certain foods, picky eating, food selectivity, sensory processing difficulties and dysphagia. The same study indicates significant feeding and swallowing difficulties in children with ASD, emphasizing the multifactorial complexity of problems related to eating and swallowing in this group. Although other people also have eating problems, the studies included in this review served to better understand aspects of eating and swallowing in the population with ASD, including swallowing assessments and individualized evidence-based interventions¹⁹.

Despite the scarcity of information about the causes of dysphagia, studies have used questionnaires to collect data from parents/caregivers about oral motor difficulties, sensory processing difficulties, and dysphagia symptoms in the ASD population.

The Brief Autism Mealtime Behavioral Inventory (BAMBI) was one of the instruments used in the studies included in this review, as an adjuvant clinical tool. The BAMBI is an observer-reported outcome measure designed specifically for parents/caregivers, created to capture specific mealtime behaviors of children with ASD and configured into questions scored on a scale of 5²⁰.

Sahan et al (2021)¹⁸ used the Karaduman Chewing Performance Scale (KCPS) as an instrument to measure the level of chewing performance

in children. The KCPS proves to be a practical resource with five different levels between 0 and 4, where higher levels indicate worse chewing performance and levels 0–2 indicate chewing function within normal limits. Additional records included in the protocol were covered, namely: oral transition time with solid foods, initial teething time and number of daily meals²¹. In the last stage of application, the priority of oral motor assessments was to observe the presence of open mouth, open bite with malocclusion, anterior propulsion of the tongue in dysfunctional movement, high arched palate and oral hygiene, recorded as ‘present’ or ‘absent’ in the questionnaire¹⁸.

The CHARGE Gastrointestinal History (GIH) protocol used by two studies^{11,14} consists of questions directed to gastrointestinal symptoms separated into 10 items per scale (0=never; 1=rarely; 2=sometimes; 3=often; 4=always). The main symptoms addressed are difficulty swallowing, abdominal pain, gas/bloating, diarrhea, constipation, pain when evacuating, vomiting, sensitivity to food, blood in the stool and blood in vomit. The questionnaire also includes four questions (yes/no) about the presence of food allergies, dietary restrictions and food aversions. Other data collected as part of the protocol includes food allergies; reasons for dietary/food restrictions; gastrointestinal diagnosis; demographic data and medications used in the last month.

Symptoms such as swallowing problems were reported after the diagnosis of gastrointestinal disorders, with the presence of behavioral changes among people with ASD. Therefore, an important topic addressed by studies is the possibility of a relationship between gastric problems and eating problems^{15,11,12,14}.

In a survey of 114 children, Ahmed and collaborators (2022)¹⁵ discovered a relationship between ASD and gastrointestinal problems. On average, more than half of children with ASD presented such symptoms, with a frequency up to four times higher than in children without ASD. Of these findings, three patients had difficulty swallowing, but did not undergo dysphagia assessment. Furthermore, it is important to highlight that symptoms such as feeding problems, dysphagia, nausea, abdominal distension, profound constipation or diarrhea may indicate problems with gastrointestinal function (oropharyngeal, esophageal and intestinal dysmotility).

It is important to highlight that the studies did not use standardized protocols in clinical assessments of dysphagia. Therefore, there is still no scientific evidence to indicate that ASD may be related to oropharyngeal and/or esophageal dysphagia. Therefore, we suggest studies on ASD and dysphagia using specific and validated protocols for a complete and effective assessment of dysphagia, in addition to the use of objective exams to assess swallowing functionality in this population.

Conclusion

The studies included in this review had as their target audience children diagnosed with ASD, who had oral motor difficulties, choking, sensory processing difficulties, vomiting, swallowing difficulties, dysphagia and gastrointestinal changes.

Identifying dysphagic symptoms in ASD can be challenging, as these symptoms may be linked to comorbidities associated with the diagnosis and behavioral patterns distinct from ASD. The results of the studies included in this review did not provide information on clinical assessments of dysphagia. Therefore, there is still no scientific evidence to indicate that ASD may be related to oropharyngeal and/or esophageal dysphagia.

Another important issue is the standardization of tools used to assess swallowing in children with ASD. The studies did not use standardized protocols aimed at assessing dysphagia. The lack of robust evidence in these studies also suggests the need for research with greater methodological rigor, appropriate and validated tools to improve the quality of evidence and to make a reliable clinical diagnosis of dysphagia in individuals with ASD.

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