

Childhood apraxia of speech and Autism Spectrum Disorder: integrative review

Apraxia de fala na infância e transtorno do espectro do autismo: revisão integrativa

Apraxia infantil del habla e Trastorno del espectro autista: revisión integradora

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Abstract

Introduction: The Disorder of the Autistic Spectrum (ASD) is a disorder of neurodevelopment characterized by persistent difficulties in social interaction and oral communication. Infantile apraxia of speech is a neurological disorder that affects who speak. There are stories that point to a relationship between disorders. Purpose: To describe the findings about Childhood Apraxia of Speech and communicative difficulties of children with ASD. Research strategy: It is an integrative revision of the literature, made under the basis of the data from Biblioteca Virtual de Saúde National Library of Medicine, COCHRANE, SCOPUS and WEB OF SCIENCE. Selection criteria: There was a selection of papers with no publishing date limit and, afterwards, there was the reading of titles and summaries. The inclusion criteria were: complete papers which addressed ASD and Apraxia of Speech, issued in Portuguese and English in national and international journals. As an exclusion criterion, the studies with children over 5 years 11 months and 29 days old, senior citizens and adolescents were not considered, which was also true for papers that did not address the Childhood Apraxia of Speech in children suffering from ASD. Data analysis: The reading of the papers was made by titles and summaries and the extraction of data to characterize the methodology and the research content. Results: There was the selection for full reading of 15 papers which had been separated according to two thematic axes

Authors' contributions:

JFS: participated in study conceptualization, data collection and analysis, manuscript writing, and approval of the final version. ACOR: participated in data analysis.

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related to ASD and Apraxia of Speech. Conclusion: The findings demonstrate the lack of consensus between the disorders.

Keywords: Apraxias; Autism Spectrum Disorder; Speech Therapy.

Resumo

Introdução: o Transtorno do Espectro do Autismo (TEA) é um transtorno do neurodesenvolvimento caracterizado por dificuldades persistentes na interação social e na comunicação oral. A Apraxia de Fala Infantil (AFI) é um distúrbio neurológico que afeta os sons da fala. Há relatos que apontam uma relação entre TEA e AFI. Objetivo: descrever os achados sobre AFI e dificuldades comunicativas em crianças com TEA. Estratégia de pesquisa: trata-se de uma revisão integrativa da literatura, realizada nas bases de dados Biblioteca Virtual de Saúde (BVS), National Library of Medicine (Medline via Pubmed), Cochrane, Scopus e Web of Science. Critérios de seleção: foram selecionados artigos sem limite de data de publicação e em seguida a leitura de títulos e resumos. Os critérios de Inclusão foram: artigos completos que abordassem Autismo e AFI, publicados em português e inglês, em periódicos nacionais e internacionais. Como critério de exclusão, foram desconsiderados os estudos com crianças acima de 5 anos, 11 meses e 29 dias, idosos e adolescentes e também artigos que não abordassem a AFI em crianças com TEA. Análise dos dados: Foi realizada a leitura dos artigos por títulos e resumos e a extração de dados para caracterizar a metodologia e o conteúdo da pesquisa. Resultados: foram selecionados 15 artigos para leitura na íntegra, os quais foram separados de acordo com dois eixos temáticos, relacionados ao TEA e AFI. Conclusão: os achados demonstram a falta de consenso na correlação entre AFI e TEA.

Palavras-chave: Apraxias; Transtorno do Espectro Autista; Fonoterapia.

Resumen

Introducción: el Trastorno del Espectro Autista (TEA) es un trastorno del neurodesarrollo caracterizado por dificultades persistentes en la interacción social y la comunicación oral. La apraxia del habla infantil (AFI) es un trastorno neurológico que afecta a los niños que hablan. Hay historias que apuntan a una relación entre TEA y AFI. Objetivo: describir los hallazgos sobre la apraxia infantil del habla y las dificultades comunicativas en niños con TEA. Estrategia de investigación: se trata de una revisión integrativa de la literatura, realizada en las bases de datos Biblioteca Virtual en Salud (BVS), Biblioteca Nacional de Medicina (Medline vía Pubmed), Cochrane, Scopus y Web of Science. Criterios de selección: se seleccionaron artículos sin límite de datos de publicación y luego se leyeron títulos y resúmenes. Se publicaron artículos de inclusión: artículos completos que abordan el Autismo y la infancia, publicados en portugués e inglés, en revistas nacionales e internacionales. Como criterios de exclusión, se descartaron estudios con niños mayores de 5 años, 1 mes y 29 días, niños y adolescentes y también artículos que no aborden la praxis del habla en el TEA. Análisis de datos: Los artículos fueron leídos por títulos y resúmenes, y el artículo de datos para caracterizar la metodología y la investigación. Resultados: Se seleccionaron 15 artículos para lectura completa, los cuales fueron separados según dos ejes temáticos, relacionados con el TEA y la apraxia del habla en la infancia. Conclusión: los hallazgos deben tener una falta de consenso entre la ocurrencia de transtornos.

Palabras clave: Apraxias; Transtorno del Espectro Autista; Logopedia.



Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by persistent difficulties in social interaction and oral communication, with persistent, repetitive, and restrictive patterns¹ and behavioral changes². It combines genetic and environmental factors, originates in the first years of life, is permanent, does not have a cure², and permanently affects the maturation of the central nervous system (CNS)³.

Some authors believe that only 10% of ASD cases are secondary to genetic disorders, chromosomal abnormalities, and serious neurological conditions^{3,4}.

In the DSM-5 classification, ASD includes Asperger syndrome, which does not have significant delays in verbal language and cognitive structures. There are signs that hinder language, but less pronounced, and behavioral differences in social aspects².

A study reports that ASD is diagnosed late due to the difficulty in finding evident signs compared to typical children before 6 months of age. From 12 to 18 months, the signs are more noticeable, favoring an earlier diagnosis, around 1 or 2 years old². Signs evident from 6 months of age help with diagnosis².

ASD is clinically diagnosed with diagnostic scales applied by specialists, without the need for additional tests to confirm the syndrome⁵.

ASD is more prevalent in male children, with a ratio of two to three boys to one girl¹⁻⁶. Also, 30% of children with ASD have intellectual disabilities, attention-deficit/hyperactivity disorder (ADHD), language deficits, depression, anxiety, neurological disorders (epilepsy), sleep disorders, genetic disorders, motor difficulties (dyspraxia, gait changes, or fine motor changes), sensory changes, genetic diseases (fragile X syndrome, tuberous sclerosis, Williams syndrome), gastrointestinal disorders, and dietary changes².

Regarding the epidemiology of ASD, the authors of studies from 2005 and 2009 report an estimated 60 children per 10,000 births in 2005 and 60 to 70 children per 10,000 births in 2009⁷⁻⁸. A more recent study in the United States shows a prevalence of 14.6 per 10,000 births⁹, whereas another one conducted by that country's Centers for Disease Control and Prevention (CDC) found a prevalence of 18.5/1,000 – i.e., one ASD case per 54 births¹⁰.

A child with ASD may have communication and language changes, sensory changes involving muscle weakness or low energy, tactile sensitivity to movement, gustatory and olfactory sensitivity, auditory and visual sensitivity, sensory search and distractibility, and less intensity in registering sensations due to hyporresponsiveness to the body, being called by name, and understanding².

In recent years, some authors³⁻⁴ began to report the relationship between ASD and child-hood apraxia of speech (CAS). However, praxic disorders are not involved in the symptomatology of children with autism; rather, they are treated as comorbidities. Praxis is the ability to contextualize, plan, and complete motor actions during the speech process¹¹. Children are not born with developed praxis; this function is learned throughout maturational development, requiring interaction with the external environment¹².

Important markers described in a study show that ASD and CAS are changes of genetic origin that compromise cognitive-linguistic abilities. Hence, they may have genes in common, such as changes in the FOXP2 gene¹³. Findings in studies reveal that about 65% of children diagnosed with ASD have CAS⁹.

According to a study, for every 1000 children between 1 and 5 years old, 10 have some communication disorder. Among these children, 3 to 5% have CAS⁷.

CAS is a neurological disorder that affects speech sounds. It hinders the precision and awareness of orofacial movements, leading to production errors (omission of sounds and syllables, distortion of sounds, substitution of sounds, and simple syllable structure) and prosody changes. It is also important to rule out muscle deficits related to such changes⁶.

The diagnosis of CAS involves some segmental and suprasegmental characteristics; when these characteristics are present in typical children, they differ from those of children with autism. Among them, the following ones can be identified: articulatory groping at the beginning of speech, substitution errors characterized by metathesis, inconsistent exchanges, and a greater number of errors in vowels. Suprasegmental characteristics refer to accents inconsistently placed on the stressed syllable and the perception of nasopharyngeal resonance¹⁴.

The criteria used in a previous study¹⁵ to diagnose CAS were the limited repertoire of consonants



and vowels; frequent omissions of phonemes; high incidence of errors in vowels; inconsistent articulation; changes in prosody, vocal quality, and fluency; difficulty imitating words, phrases, and syllabic structures; abnormal voluntary oral movements; reduced language expressions; and reduced diadochokinetic skills.

There is no consensus on the number of criteria to define the diagnosis. Some authors state that more than five criteria are necessary, while others report at least eight¹⁴. This study aimed to analyze the occurrence of CAS with ASD.

Research strategy

This integrative review of the literature was developed based on national¹⁶ and international recommendations¹⁷. Its research question was, "What is the relationship between CAS and ASD, considering diagnostic and therapeutic approaches?".

After defining the research question, the literature was searched in stages. The researchers initially delimited the research problem, defined the databases and other sources of information to

search for studies, planned and developed search strategies, recorded the search, evaluated the results, reported the search and selection process, and evaluated and synthesized the findings.

The research question was defined by surveying key concepts related to the central objective of the research, namely ASD and CAS. The bibliographic databases selected were LILACS via the Regional Portal of the Virtual Health Library (VHL), MEDLINE via PubMed, and CINAHL, Cochrane, Scopus, Web of Science, and PsycINFO via the Capes Journal Portal, filtered for Portuguese and English publications. All original and review articles were consulted without excluding the year of publication to collect data and changes over time. Duplicate references were excluded. The search strategy was planned to find studies that were at the intersection of the sets that contain the key concepts, using the Boolean operators in Portuguese and English that expand the scope of the search and connect the concepts to refine the search (Chart 1).

Chart 1. Search strategy

Database	Strategy/Formula		
VHL Regional Portal	(apraxias OR dispraxia OR "Apraxia, Ideomotor" OR "Apraxia Ideomotora" OR "Dispraxia Ideomotora" OR "Speech Disorders" OR "Trastornos del Habla" OR "Distúrbios da Fala" OR aprosodia OR "Apraxia de Fala" OR "Apraxia de Fala" on aprosodia OR "Apraxia de Fala" OR "Apraxia de Fala" OR "Childhood Apraxia of Speech") AND ("Autism Spectrum Disorder" OR "Trastorno del Espectro Autista" OR "Transtorno de Espectro Autista" OR "Transtorno de Espectro do Autismo" OR "Autistic Disorder" OR "Trastorno Autístico" OR "Transtorno Autístico" OR "Autismo Infantil" OR "Síndrome de Kanner") AND (db:("LILACS" OR "IBECS" OR "INDEXPSI" OR "BINACIS" OR "LIS"))		
MEDLINE via PubMed	(Apraxias OR "Apraxia, Ideomotor" OR "Speech Disorders" OR "Apraxia of Speech" OR "Childhood Apraxia of Speech") AND ("Autism Spectrum Disorder" OR "Autistic Disorder")		
Cochrane	(4		
Scopus	(Apraxias OR "Apraxia, Ideomotor" OR "Speech Disorders" OR "Apraxia of Speech" OR "Childhood Apraxia of Speech") AND ("Autism Spectrum Disorder" OR "Autistic Disorder")		
Web of Science	C		



Selection criteria

The inclusion criteria were articles published in full, in Portuguese and English, which addressed CAS and ASD in children aged 1 year, 11 months, and 29 days to 5 years, 11 months, and 29 days diagnosed with ASD, in the age range of oral language development. Exclusion criteria were articles that addressed apraxia of speech and ASD in adults and older adults and CAS in children without a diagnosis of ASD.

Data analysis

The study began identification by reading the titles and abstracts of the selected articles, based on the selection criteria (articles that referred to CAS and ASD in children aged 1 year, 11 months, and 29 days to 5 years, 11 months, and 29 days. Altogether,

1,363 articles were found – 26 articles via VHL, 199 articles via PubMed, and 1,138 via the Capes Journal Portal. After inspecting these articles, 562 duplicates were excluded, as they appeared in more than one database. After reading the titles of 801 articles, 531 were excluded because they did not make any reference to the study topic. Thus, 270 articles were selected for abstract reading – 246 of them were excluded according to the exclusion criteria, and 24 articles were selected to be read in full. After reading their full text, nine did not address CAS and ASD and were excluded. Therefore, content citation records included the remaining 15 articles, of which nine were national and six were international (Figure 1).

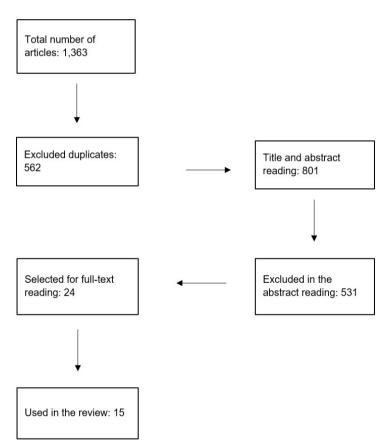


Figure 1. Flowchart with the article selection process for the literature review



Results

Most of the included articles were systematic literature reviews. They were distributed as follows: six systematic reviews, four case studies, two dissertations, and three integrative review articles. The second most frequent methodological design among the 15 articles found was the case study (Chart 2).

Chart 2. Articles selected for the review

Authors and year	Country	Study type	Objective	Main Findings
Rubem Abraão, Simone Herrero, Luciana Paula de Vitto, 2007.	Brazil	Case study	To describe the speech- language-hearing therapy intervention in a 24-month-old male child.	It describes the 9-month intervention and the significant evolution in the therapeutic process.
Thais Souza and Luzia Payão, 2008.	Brazil	Bibliographic review	To differentiate childhood apraxia of speech (CAS) from language disorders.	It investigates the similarities and differences between CAS and other language changes.
Micheline Silva, James Mulick, 2009.	USA	Bibliographic review	To identify comorbidities, etiologies, and incidences of possible changes in children with autism spectrum disorder (ASD).	It used assessment tools to assist in the diagnosis of language changes.
Lawrence Shriberg et al., 2010.	USA	Literature review	To analyze the treatment of children aged 4 to 5 years with ASD and CAS.	It describes the importance of early intervention after the diagnosis of CAS and associated ASD and its possible hypotheses.
Cheryl Tierney, Marie Kurtz, and Heather Souders, 2012.	USA	Case study	To correlate ASD with CAS and their findings.	It related CAS to ASD, a new view on the relationship between these disorders.
Cheryl Tierney et al., 2015.	USA	Bibliographic review	To report the relationship between CAS and comorbidities in children with ASD.	Study in autistic children who underwent an assessment with a diagnosis of CAS.
Marília Berbal, 2018.	Brazil	Dissertation (thesis)	To describe possible changes in children with ASD and CAS.	Study in autistic children to analyze the possible diagnosis of CAS.
Fernanda Martins, 2018.	Brazil	Case study	To analyze the praxis of four children with ASD.	It correlated the findings between CAS and ASD.
Eugênio Conti, et al., 2020.	USA	Bibliographic review	To describe the relationship of children with ASD who present language disorders and CAS.	The study shows that 98% of children with ASD who were evaluated and diagnosed with CAS improved after the intervention.
Karen Chenausky et al., 2020.	USA	Bibliographic review	To Investigate the factors related to CAS and ASD.	The study shows the relationship between CAS and ASD.
Aline Oliveira, Layla Marcelino, Alexsandra Ferreira, 2020.	Brazil	Systematic review	To Investigate the factors related to CAS and ASD.	It investigates the factors related to CAS and ASD in children under 5 years old.



Authors and year	Country	Study type	Objective	Main Findings
Cíntia Braz et al. 2020.	Brazil	Systematic review	To investigate signs of risk for CAS in children aged 6 to 24 months.	The study describes signs that point to evidence for CAS and its relationship with other disorders.
Francisco Assumpção and Ana Pimentel, 2020.	Brazil	Case study	To associate the etiology with the clinical condition of children with ASD and their language delays.	It relates ASD to a cognitive deficit, seeking etiologies and prognoses after interventions established by the team.
Fernanda Martins et al., 2021.	Brazil	Case study	To report the findings in children aged 3 to 5 years diagnosed with ASD with CAS.	It relates CAS to ASD changes.
Karina Homem, 2021.	Brazil	Literature review	To search for clinical findings that correlate and differentiate CAS and ASD.	It describes the CAS and ASD clinical findings.

Discussion

The predominance of the scientific production in question addressed the identification or reports of CAS in ASD¹⁹⁻²⁰. However, a study shows that children with CAS, as well as children with ASD, have communication difficulties related to speech delay²¹. Both may have isolated speech changes, be associated with other similar disorders, or be linked in the same diagnosis²².

Both disorders have some differences and similarities²³. However, the lack of instruments that bring together adequate characteristics that lead to a differential diagnosis requires mastery in protocol application, more structured knowledge, management in test application, and observation of signs and symptoms²⁴.

CAS is a change in speech motor planning observable in children under 1 year old, when some relevant signs are already present, such as reduced babbling and unusual quietness, despite their good social interaction. After 12 months old, the child tends to become more irritable because they speak little. They have good comprehension, but the first words may appear around 48 months of age, using facial expressions, gestures, and nonverbal sounds as a reference to communicate. They emit isolated sounds, such as vowels instead of consonants, and produce isolated syllables to communicate¹⁵.

Children with CAS have adequate communicative intention, but they cannot properly plan structures and program the sequence of speech coarticulation movements, which leads to slow, paused speech and prosody difficulties. These are added to sensory changes that involve activities of daily living (ADLs) and food selectivity, affecting swallowing and orofacial movements with the tongue, such as blowing and sucking²⁵.

A study indicates that CAS and ASD are correlated, with CAS manifestations being more pronounced in children with ASD than in children without neurodevelopmental disorders⁴. Despite the agreement that these two conditions are correlated, studies on the topic still lack a consensus. This was mentioned in another study, which states that this relationship is not that frequent, since children with autism have a delay in social reciprocity due to neurodevelopmental disorders, leading to a limitation in the structure of the means of communication, making it more difficult, and thus confusing it with the diagnosis of CAS²⁶.

A study reports a divergence between both conditions, as in CAS there is no absence of eye contact, whereas in ASD it is avoided. This changes socio-communicative skills, due to the absent or decreased visual search²⁰.

Some authors in previous studies found a significant relationship between CAS and ASD in expressive language, with greater losses in oral motor skills, prosody, oral praxis, and oral speech sound production. Hence, further studies are needed to improve the prognosis and aim for more accurate and effective speech-language-hearing therapy interventions⁴⁻¹⁵⁻²⁰⁻²⁷.



Furthermore, the study states that some children with ASD showed deficits in social communication and social interaction and changes in orofacial motor imitation, similar to those children with suspected CAS²⁸.

Some authors argue that both CAS and ASD impair verbal motor performance, due to neuronal issues involving mirror neurons and the frontal lobe. However, there is a need for more in-depth studies²⁷⁻²⁸.

Conclusion

The national and international literature listed on the topic was relevant, as some studies reported a significant relationship between CAS and ASD. However, not all findings lead to the conclusion that this double occurrence will be found in all children with autism. Other studies do not corroborate this relationship since the considerations on the topic were more related to signs of ASD than to CAS.

There are different approaches to diagnosing CAS in children with ASD and different assessment methods to analyze oral and verbal praxis, which is why studies correlating CAS and ASD are difficult to find.

Another justification is the lack of a validated Brazilian protocol, which could lead to false positives during the diagnosis. Thus, greater investment in national studies is suggested to increase research on the topic to seek more assertively the diagnosis of CAS and its relationship with ASD.

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