

Review Article

Selective Mutism and Autism Spectrum Disorder: A Systematic Review

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ABSTRACT

Autism is characterized by disturbances in communication, behavior, and social interaction, where repetitive and restricted interests predominate. Selective mutism (SM) and autism often coexist; however, the relationship between them is complex and not yet fully understood. This systematic review aimed to analyze research on the comorbidity between autism and SM, identify trends in the field, and highlight areas for future research. Following the PRISMA methodology, eight studies met the inclusion criteria. The results indicate that many children with autism also present with selective mutism, with the onset of symptoms occurring later than in children without autism. From the parents' perspective, caring for a child with both SM and autism is challenging, as it exacerbates parental stress and negatively impacts the overall quality of life for the family. Various treatments, including pharmacological interventions, music therapy, and behavioral programs, have been shown to help reduce the symptoms of selective mutism in children and adolescents diagnosed with autism. In conclusion, the findings of this review provide an up-to-date foundation for understanding the comorbidity of SM and autism. Furthermore, it offers guidance for health and education professionals to expand their knowledge of SM and its relationship with autism, facilitating a better understanding of the interplay between these two diagnoses.

Keywords:

Autism; Autism Spectrum Disorder (ASD); Selective Mutism (SM); Anxiety Disorder

Mutismo Selectivo en el Trastorno del Espectro Autista: una revisión sistemática

RESUMEN

En el autismo existen alteraciones en la comunicación, la conducta y la interacción social, predominando intereses repetitivos y restringidos. El mutismo selectivo (MS) y el autismo pueden ser comórbido en muchas ocasiones, pero esta relación es compleja y aun no se comprende bien. El objetivo de esta revisión sistemática fue analizar las investigaciones que estudian la comorbilidad entre el autismo y el MS, delinear tendencias en el campo e identificar áreas para futuros estudios. Se utilizó como guía la metodología PRISMA y 8 estudios cumplieron con los criterios de inclusión. Los resultados revelan que muchos niños/as con autismo también presentan MS, siendo la aparición de los síntomas más tardía en comparación a grupos que no presentan autismo. Desde la perspectiva de los padres, existen complejidades para cuidar a un niño con MS y autismo, ya que esta tarea exacerba el estrés de los padres y afecta la calidad de vida de la familia en general. Existen diversos tratamientos como los farmacológicos, musicoterapia, programas y/o estrategias conductuales, que ayudan a disminuir los síntomas de MS en niños/as y adolescentes diagnosticados con autismo. Se concluye que los resultados de esta revisión sistemática pudiesen servir de base para comprender desde una perspectiva actualizada la comorbilidad del MS y el autismo. A su vez permite a los diferentes profesionales de la salud y/o educación prepararse y expandir los conocimientos acerca del MS y su relación con el autismo, orientando a los diferentes profesionales de la salud respecto de la relación entre ambos diagnósticos.

Palabras clave:

Autismo; Trastorno del Espectro Autista; Mutismo Selectivo (MS); Trastorno de Ansiedad

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Received: 10-09-2023

Accepted: 01-06-2025

Published: 04-15-2025

INTRODUCTION

Autism Spectrum Disorder (hereinafter referred to as autism or ASD) is a neurodevelopmental disorder of neurobiological origin, which begins in childhood. It affects the development of communication, social interaction, and behavior, presenting with restricted and repetitive behaviors and interests (American Psychiatric Association [APA], 2013). Diagnosis can be made at an early age, which is crucial for promoting autonomy and socialization, as well as helping the family prepare (Thompson-Hodgetts et al., 2020). The prevalence of autism has increased exponentially in recent decades and varies both regionally and internationally, with a median of 100 cases per 10,000 people (range: 1.09 per 10,000 to 436.0 per 10,000) (Zeidan et al., 2022). Clinical practice has shown that autistic children and adolescents additionally experience a wide range of communication and anxiety disorders. Moreover, they may present speech sound disorders (Peter et al., 2019), language disorders (Andreou et al., 2022; Georgiou & Spanoudis, 2021), and selective mutism (hereinafter SM) (Panasiuk, 2019). Often, the presence of SM is obscured by other symptoms characteristic of autism, preventing it from being considered a comorbid psychiatric condition. Early diagnosis is essential, but the semiological boundaries between SM and autism are often unclear for medical professionals, making it a topic of ongoing debate and study (Steffenburg et al., 2018).

According to the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), SM is an anxiety disorder characterized by a consistent inability to speak in specific social situations where this is expected, despite the ability to do so in other situations (APA, 2013). Symptoms of SM emerge in childhood, and its prevalence ranges from 0.1% to 2.2%, with a higher incidence in girls than in boys (Koskela et al., 2023; Oerbeck et al., 2018). Although this condition is more commonly described in children, it can also be observed in adults (Muris & Ollendick, 2021). Communication patterns in SM vary from a total absence of speech in almost all social situations to the absence of speech only in certain situations (Oerbeck et al., 2018). The onset of symptoms often occurs in school, where social demands are higher. In this context, the child is less likely to speak with the teacher, may speak only to some peers, or may not communicate with anyone at all (Driessen et al., 2020). Generally, children speak normally at home with their families; however, some may talk at school but not at home (Steffenburg et al., 2018).

The clinical overlap between SM and autism is receiving increased attention, as 62.9% of children diagnosed with SM may also have an ASD diagnosis (Muris & Ollendick, 2021;

Steffenburg et al., 2018). This can be explained by the social anxiety associated with autism (Settipani et al., 2014; Sharkey & McNicholas, 2008) as well as speech and language difficulties (Cohan et al., 2008; Müller et al., 2008). Recent research has shown that many children diagnosed with SM also exhibit pathognomonic features of autism. For example, a study by Kearney & Rede (2021) found that a high percentage of children with SM experience clinical symptoms of anxiety and developmental disorders, suggesting a correlation with autism. A study by Steffenburg et al. (2018), conducted on a sample of 71 girls and 26 boys, also revealed that more than 50% met the criteria for ASD.

Both disorders can involve communication difficulties, and it is crucial to note they are independent of each other and have different origins. Causes of SM are typically related to anxiety, whereas autism is linked to the pathognomonic neurological characteristics of individuals. The similarity in social and sensory symptoms in both disorders could explain the difficulty in differentiating them. This often hinders adequate interventions at key developmental stages. Additionally, the notable feature of silence in SM may divert clinicians' attention, causing them to overlook other characteristic signs of autism.

The prevalence of autism and SM remains poorly understood, which has led many researchers and clinicians to overlook the co-occurrence of both conditions and their consequences on socialization and quality of life (Simms, 2017). Much of the literature on SM and autism comorbidity struggles to establish clear diagnoses. This is largely due to the frequent exclusion of people with severe forms of autism, which limits a comprehensive understanding of the relationship. Moreover, the core behaviors of autism, like socialization difficulties and reduced social motivation, can sometimes overshadow SM-related behaviors in certain children (Cholemkery et al., 2014). Despite the complex prevalence and growing clinical interest in this comorbidity, to our knowledge, no systematic review has yet been conducted to explore SM in people with autism.

This systematic review aimed to analyze existing research on the comorbidity between autism and SM, outline emerging trends in the field, and identify areas for future investigation. Specifically, the following research questions were posed: 1) What are the characteristics and findings of studies on SM and autism? 2) What methods have been used to assess SM in people with autism? 3) What is the quality of the existing research on SM and autism?

The findings of this review will contribute to an up-to-date understanding of the comorbidity between SM and autism. Furthermore, they will provide valuable insights for health and education professionals—such as physicians, psychologists, psychiatrists, and others—helping them to better prepare for and expand their knowledge about SM and its relation to autism, and to approach the condition with greater clarity.

METHOD

Protocol and Registration

The review was registered online in the International Prospective Register of Systematic Reviews (PROSPERO; CRD42022382692), and it adhered to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Page et al., 2021).

Eligibility Criteria

PRISMA guidelines require clear inclusion and exclusion criteria to ensure relevant and specific results (Page et al., 2021). Studies were included if they met the following criteria: (1) peer-reviewed empirical research articles (quantitative, qualitative, or mixed-methods) published in English, indexed in Scopus, PubMed, Web of Science, or EBSCOhost from database inception to the present; and (2) studies examining the relationship between Selective Mutism (SM) and autism. The following were excluded: (1) systematic reviews, meta-analyses, umbrella reviews, opinion pieces, narrative reviews, dissertations, editorials, book chapters, grey literature (e.g., doctoral theses), and conference proceedings; and (2) articles published in languages other than English.

Search Strategy

In December 2022, we searched for studies examining the relationship between SM and autism, using the databases Scopus, PubMed, Web of Science, and EBSCOhost. A second search was conducted in January 2023 using Google Scholar to update the initial results. Filtering tools were applied to include only peer-reviewed journal articles, with double-blind review, written in English, and published between 1976 and 2023. Search terms combined keywords using Boolean operators: (autism* OR autistic OR Asperger* OR autism spectrum disorders*) AND (selective mutism OR elective mutism*)*. Database-specific search field tags were used—for example, *TITLE-ABS-KEY* in Scopus, *TS* in Web of Science, and *Title/Abstract* in PubMed.

Study Selection

After compiling the articles retrieved from all databases, duplicates were manually removed ($n = 103$). Two authors independently and simultaneously screened the titles and abstracts of the remaining 94 articles. Reports considered potentially relevant by both reviewers and meeting the inclusion criteria were subjected to full-text screening. A snowballing method was also employed to manually identify additional articles from the reference lists of included studies. The newly identified articles were independently assessed again through full-text review. Inter-rater reliability was calculated using Cohen's Kappa ($\kappa = 0.85$), and discrepancies were resolved through a Zoom meeting. Finally, 8 studies were included in the qualitative synthesis (see Figure 1).

Data Extraction

A semi-structured, pre-designed Excel spreadsheet was used for data extraction. This allowed us to systematically organize the information extracted from the studies, facilitating both synthesis and analysis of the results. For each article, the following data were extracted: information about the research team (including location); study details (type of design); participant characteristics (sample size, age, gender); methods (scales/subscales, informants, and reported psychometric properties); and main findings related to Selective Mutism and comorbid autism. Before data extraction, three of the authors discussed each article to agree on relevant elements, in order to avoid misinterpretations or omissions. Subsequently, two authors independently and in parallel extracted the data from all included articles. Discrepancies in the extracted data were resolved through five online meetings via Zoom, resulting in 100% agreement. Finally, the data from all included articles were summarized using a narrative synthesis approach.

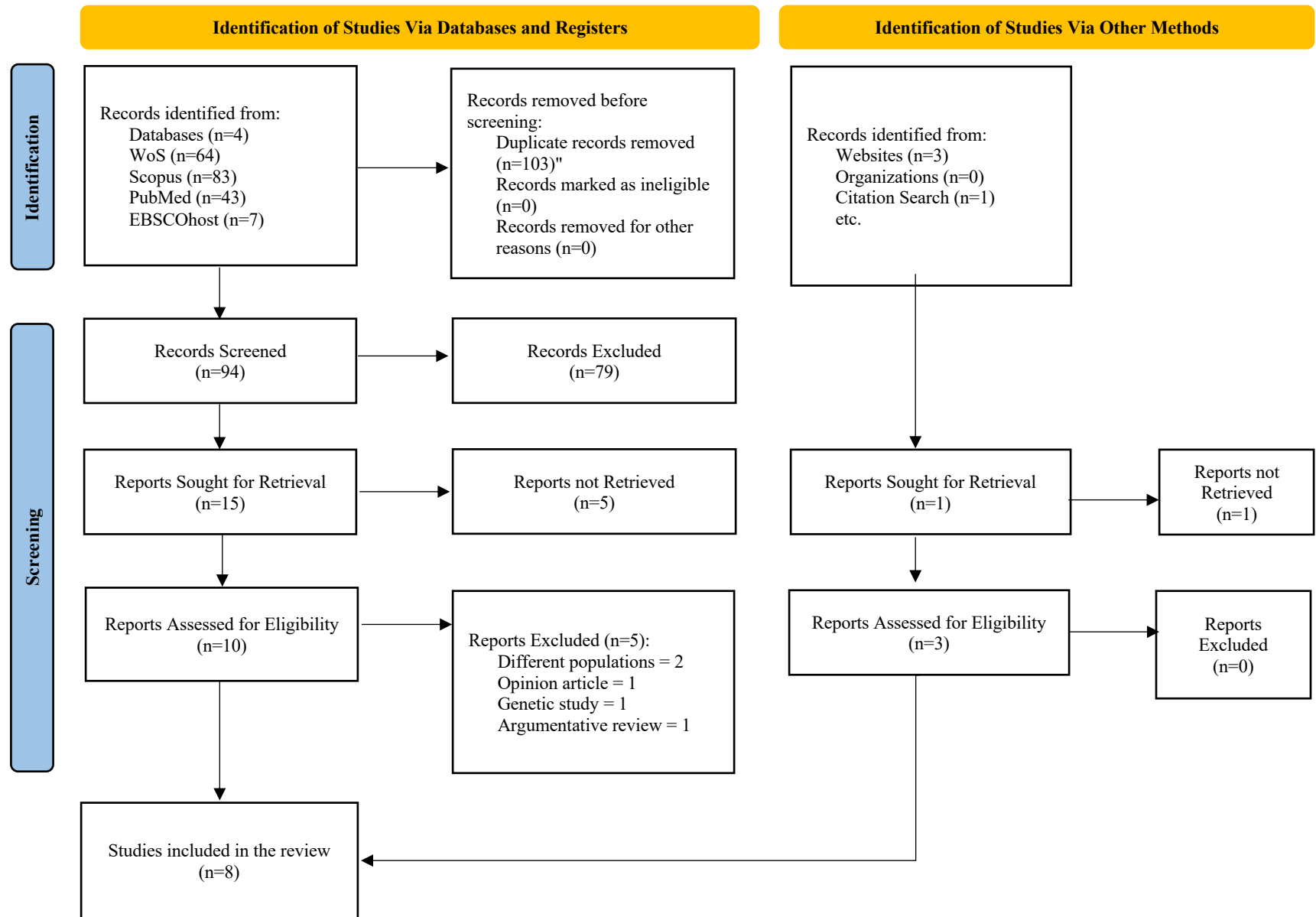


Figure 1. Flow Diagram.

Quality Assessment

The quality of the included articles was assessed by the lead researchers, and any uncertainties were resolved through discussion with the full research team. The Quality Assessment Tool for Studies with Diverse Designs (QATSDD; Sirriyeh et al., 2012). was used. This tool has been previously employed in various systematic reviews related to the autism spectrum (Adams & Young, 2021; Goswami et al., 2024). QATSDD consists of 16 items and is suitable for evaluating the quality of quantitative, qualitative, and mixed-methods research. Fourteen of the criteria apply to qualitative studies, with a maximum possible score of 42, and the same applies to quantitative studies. All 16 criteria apply to mixed-methods studies, with a total possible score of 48. Each item is rated on a scale from 0 to 3, with higher scores indicating higher methodological quality. Any items that raised concerns for the lead researcher were discussed collaboratively with the other authors.

RESULTS

Studies Included

Eight articles met the inclusion criteria. Detailed information for each study is provided in Appendix 1. All studies were published within the last five years and conducted across six countries: the United Kingdom (n = 3), the United States (n = 2), Sweden (n = 1), India (n = 1), Australia (n = 1), and South Africa (n = 1). The journals covered a range of research topics, including the treatment and intervention of SM and autism (n = 3 articles), the onset and symptomatic features of SM (n = 3 articles), and the correlation between SM and autism (n = 2 articles). The articles were published in the following journals: *Clinical Case Studies* (n = 1), *Neuropsychiatric Disease and Treatment* (n = 1), *Frontiers in Psychology* (n = 1), *Asian Journal of Psychiatry* (n = 1), *Journal of Autism and Developmental Disorders* (n = 1), *International Journal of Developmental Disabilities* (n = 1), *Child Psychiatry & Human Development* (n = 1), and *Australian Journal of Music Therapy* (n = 1).

Methodologies

Of the eight studies reviewed, four employed quantitative methods (Ludlow et al., 2022; Muris et al., 2021, 2023; Steffenburg et al., 2018), three studies adopted qualitative approaches (Keville et al., 2023; Valaparla et al., 2018; Wakamatsu, 2022), and only one study employed a mixed-methods design (Cengher et al., 2021).

Data Collection Methods

The studies used various tools to assess SM and autism. These included: the Selective Mutism Questionnaire (SMQ; Bergman et al., 2008), the Preschool Anxiety Scale-Revised (PAS-R; Edwards et al., 2010), the Autism Spectrum Questionnaire (ASQ; van der Ploeg & Scholte, 2014), the Short Form of the Behavioral Inhibition Questionnaire (BIQ-SF; Edwards, 2007), the Autism Spectrum Screening Questionnaire (ASSQ; Ehlers et al., 1999), the Revised Child Anxiety and Depression Scale—Parent Report (RCADS-P; Chorpita et al., 2000), and the Sensory Profile 2 (SP2; Dunn, 2014). Cognitive abilities were assessed using the Wechsler Preschool and Primary Scale of Intelligence (Wechsler, 2005), the Wechsler Intelligence Scale for Children (Wechsler, 2005), and the Wechsler Adult Intelligence Scale (Ryan et al., 2003). For children with cognitive ages below the minimum requirements of the Wechsler scales, the Griffiths Developmental Scales I and/or II (Alin-Åkerman & Nordberg, 1980) were administered. Furthermore, data collection included semi-structured interviews with parents (Cengher et al., 2021; Keville et al., 2023), custom-made questionnaires (Muris et al., 2023), and medical file reviews (Valaparla et al., 2018; Wakamatsu, 2022).

Quality Assessment

The QATSDD ratings reflected the methodological strengths and weaknesses of the studies included in the review. The highest quality scores were obtained by Muris et al. (2023) and Ludlow et al. (2022). Conversely, methodological weaknesses were concentrated on three specific items. The lowest scores were related to the description of data collection procedures—only the study by Steffenburg et al. (2018) adequately addressed this component. Additional low-scoring items pertained to sample selection and its representativeness concerning the target population. The studies by Valaparla et al. (2018) and Cengher et al. (2021) received the lowest scores in this domain (see Appendix 2).

Characteristics of the Participants

There were 663 participants across all the studies analyzed, 211 of which were girls (31.8%), 188 boys (28.4%), and 264 parents (39.8%). Percentages were obtained manually through Excel, by adding the number of participants in each study and using the appropriate formula, based on the total participant number. The ages of the pediatric patients ranged from 5 to 18 years. The studies employing different informants such as children and parents as their recruitment method had a bigger sample size (Keville et al., 2023; Ludlow et al., 2022; Muris et al., 2021,

2023; Steffenburg et al., 2018). On the other hand, the samples based on clinical cases without multiple informants were smaller (Cengher et al., 2021; Valaparla et al., 2018; Wakamatsu, 2022).

Main Findings

Due to the diversity of findings across the eight studies, results were categorized into three themes: (1) Prevalence and Symptoms, (2) Parental Experiences, and (3) Interventions.

1. Prevalence and Symptoms

We identified five studies that examined the prevalence and comorbid symptomatology of autism and SM (Cengher et al., 2021; Ludlow et al., 2022; Muris et al., 2021, 2023; Steffenburg et al., 2018). One of them, Cengher et al. (2021), conducted a case study to assess the functions of selective mutism. The authors identified SM as a strategy used by autistic children and adolescents to avoid social interaction, particularly in the presence of unfamiliar individuals. Ludlow et al. (2022) explored the different factors contributing to the development of social anxiety in children with SM, with and without autism. Their findings showed that the group with both SM and autism had significantly higher levels of social anxiety and sensory avoidance compared to the group with SM only. They also tested a simple mediation model, which highlighted that sensory avoidance mediated the relationship between the diagnosis and social anxiety. Similarly, Muris et al. (2021) examined the relationships between SM, social anxiety, autistic traits, and behavioral inhibition to the unfamiliar (i.e., the tendency to respond with restraint and withdrawal when faced with novel stimuli and situations). Their results revealed positive correlations between SM, social anxiety, and autistic features, as well as a connection with behavioral inhibition—suggesting that both social anxiety and autism influence non-speaking behavior. In a related study, Muris et al. (2023) investigated the psychopathological and temperamental correlates of SM symptoms in a mixed sample of non-clinical and clinically referred patients (including people with autism). The findings indicated that SM symptoms were strongly associated with social anxiety and an anxiety-prone temperament (behavioral inhibition) and that autistic traits play a role in the selective speaking behavior observed in children. Finally, Steffenburg et al. (2018) examined the prevalence of autism in a clinical sample of children with SM. Their findings revealed that SM and autism are highly comorbid. The SM+autism group showed a later onset of symptoms, older age at diagnosis, a more frequent history of speech delay, and a higher proportion of borderline intellectual functioning or intellectual disability.

2. Parental Experiences

We identified three studies focusing on the perspectives and experiences of parents of children with SM and autism (Cengher et al., 2021; Keville et al., 2023; Ludlow et al., 2022; Muris et al., 2021). Keville et al. (2023) conducted a qualitative study in which they interviewed eleven mothers and one father of children with SM and autism, aged between 5 and 18 years. The transcript analysis revealed the following themes: 1) the complexities of concurrent issues, 2) the overwhelming impact of selective mutism, 3) the diagnostic journey, and 4) the search for solutions and support. Judgments and minimization of symptoms by educational and health systems prolonged the diagnostic process, hindering appropriate interventions. Caring for children with SM and autism, along with broader misunderstandings, increased parental stress and impacted family life. Parental advocacy and safe environments made it easier to manage contextual mutism in children. On the other hand, Cengher et al. (2021) explored the family and individual history of children diagnosed with autism and SM, through parental experiences. Notably, those who implemented treatments in naturalistic settings allowed the effects to generalize to other environments. It was found that SM symptoms were associated with higher levels of autistic traits. Furthermore, comparing correlation coefficients revealed that the relationship between SM and social anxiety (SA) was significantly stronger than the association between SM and autistic traits, with a Z value of 3.97 and $p < 0.001$ (Muris et al., 2021). Another study conducted by Ludlow et al. (2022) considering parents' opinions found that the group of participants with SM and autism exhibited significantly higher levels of social anxiety and sensory avoidance compared to the group with only SM (Ludlow et al., 2022).

3. Interventions

Two types of interventions were identified: multimodal and unimodal.

Multimodal Interventions

According to the criteria established by Cengher et al., (2021), children diagnosed with SM and autism may be more resistant to multidisciplinary treatment or may require a higher intervention dosage to achieve clinically significant outcomes (Cengher et al., 2021). In this context, Valaparla et al. (2018) implemented a multimodal intervention that combined pharmacological and non-pharmacological strategies for a patient with SM and autism. The authors found that selective serotonin reuptake inhibitors, such as fluoxetine, were effective within a therapeutic model incorporating techniques such as systematic desensitization,

modeling, token economies, and speech therapy techniques to improve voice volume. Following a 60-day hospitalization period, the patient showed significant improvement in her communication patterns, starting to interact verbally with strangers in the presence of her mother. Three months after discharge, her family reported that she had stopped communicating with gestures and was speaking openly with friends and teachers at school. This study also showed that active family involvement, along with the therapy team and school staff, creates a multidisciplinary strategy that can lead to better outcomes in the disruptive behaviors of people with autism and SM.

Unimodal Interventions: Music Therapy

Music therapy was the only unimodal intervention identified in this review. A case study was conducted with an adolescent diagnosed with SM and autism, who exhibited significant social anxiety and communicated exclusively in Vietnamese with her family. Initially, music therapy sessions were held in the clinic, and later, they were moved to the patient's home due to her anxiety. These sessions focused on singing with karaoke in Vietnamese to facilitate her participation, which later evolved to include songs in English (with and without lyrics) and incorporated functional phrases. The intervention was optimized through interdisciplinary collaboration and teletherapy during the pandemic. After four years, the patient progressed from being nearly non-verbal to singing in two languages and performing in public, showing greater confidence and verbal communication. These improvements were attributed to music therapy and a multidisciplinary approach that reduced her anxiety. The findings suggest that music therapy is a favorable alternative in SM and autism intervention, particularly at early ages and over shorter periods, fostering interaction with others (Wakamatsu, 2022). However, these results are based on a case study, and it is necessary to replicate this intervention in larger samples.

DISCUSSION

This study aimed to analyze research addressing the comorbidity between autism and selective mutism (SM), outline trends in the field, and identify areas for future studies. The findings indicate limited information on SM and autism, and therefore, this relationship should continue to be studied to achieve a more comprehensive understanding of the phenomenon.

One relevant finding is that the onset of SM symptoms occurs later in children who experience both autism and SM compared to

children who only present with SM. A possible explanation could be that this coexistence may complicate the early identification of SM-related issues. Since children with autism can experience difficulties in communication or social interaction, their SM symptoms might go unnoticed or be misinterpreted by clinicians and other socializing agents, such as teachers (Anderson, 2018; Driessen et al., 2020; Fortea Sevilla et al., 2016).

Another key result is that the comorbidity of SM and autism in children affects all aspects of life, including the ability to form friendships, find their voice, and participate in school and cultural activities (Keville et al., 2023; Muris et al., 2021, 2023). This is consistent with other studies that have explored anxiety as a trait in autistic patients, where higher levels of anxiety are associated with greater irritability and difficulties in recognizing emotions and responding socially (Jolliffe et al., 2022). Therefore, the ability to adapt and develop functional resources in response to new demands may trigger cognitive and physical symptoms of anxiety (Falconi et al., 2018).

There were also relevant findings regarding the parents' perspective, especially regarding the complexities involved in caring for a child with SM and autism, which increases parental stress and affects the family. Consequently, it is crucial to consider parental care and ensure safe environments that provide better opportunities for children to manage SM (Keville et al., 2023). This aligns with a study on parents and the approach to anxiety and autism, which asserts that anxiety is related to coping styles focused on both problem-solving and emotions. These approaches aim to reduce distress and pain when raising a child diagnosed with autism, seek actions to modify the conflict and reduce the emotional impact of the challenging situation (Canseco & Vargas, 2020). Another reason why the comorbidity of SM and autism impacts parental well-being and social development is that parents must deal with multiple challenges and difficulties during the diagnostic process, including a lack of understanding and misinterpretations by healthcare and educational systems. These factors increase parental stress and reduce their quality of life (Keville et al., 2023). Similarly, other studies indicate that parents of children with autism and other comorbid disorders experience higher levels of caregiving burden, which affects various aspects of functioning, including physical, emotional, social, and cognitive domains (Patel et al., 2022; Roper et al., 2014).

Additionally, this review found data about interventions for SM and autism. Several therapeutic modalities (pharmacological, music therapy, and behavioral programs and/or strategies) were identified, which reduced anxiety, promoted communication, and helped generalize speech (Valaparla et al., 2018). Despite the

effectiveness of these behavioral interventions, they require good adherence to treatment to achieve permanent behavioral change (Cengher et al., 2021). Research on the treatment of anxiety disorders in people with autism concurs with the use of both pharmacological and psychological approaches. However, few clinical trials prove their efficacy and safety at the pharmacological level. On the other hand, cognitive-behavioral therapy has proven effective for various mental health disorders.

Although there is a wide range of research related to anxiety in autism, there is a lack of studies exploring the relationship between SM and autism. Such research would help determine how the combination of SM and autism affects the sociocultural development of people with both disorders. There may be many reasons for the lack of research in this area, such as the absence of specialized measures for SM and ASD, which is common in anxiety disorders. Another reason is the barriers encountered in conducting research, namely the lack of representative samples of people with a formal diagnosis of comorbid SM and autism.

Areas for Future Research

Further research is needed to incorporate well-validated, specific measures of SM that can be completed by multiple informants and are suitable for use with children on the autism spectrum. Future work should employ various methodological approaches that allow for the triangulation of information. This is crucial to achieve a comprehensive perspective of the issues associated with the comorbidity of SM and autism. No studies were found specifically addressing the opinions and experiences of teachers regarding children with SM and autism, highlighting the urgent need to investigate this area.

Limitations

One limitation of this study is that it only includes articles in English. While this is the most widely used language in scientific publications, there may be literature in other languages that provides additional information on the topic. Thus, it is recommended to replicate this review in different languages. Another limitation is that, although relevant databases addressing autism and SM research such as Scopus, PubMed, Web of Science, and EBSCOhost, were used, more localized databases such as Scielo were not included. Sources like the latter would provide a perspective that considers the Ibero-American context.

CONCLUSIONS

The characteristics of autism are often overshadowed by the symptoms of selective mutism because difficulties in socialization and social motivation can make the symptoms of autism less visible. The involvement of parents in early diagnosis is essential for effective treatment and prognosis, promoting communicative and social development. Regarding intervention, cognitive-behavioral therapy has been shown to be effective, as it focuses on developing skills and finding new strategies to manage the difficulties faced by people with autism. Currently, there is more research addressing both SM and autism, indicating a growing interest in this area. However, further studies are needed, as research remains limited. This systematic review could encourage future research on adolescents, adults, and older adults, populations in whom SM has not been sufficiently explored.

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APPENDIX 1. Studies Included in the Review.

Study	Country	Objective	Design	Sample	Data Collection Methods	Main Results	Journal
Cengher et al. (2021)	USA	Assessing the functions of selective mutism	Mixed	Autistic girl, 11 years old, with ADHD, ASD, SM Julie's biological mother and father and her step-mother Staff from Julie's school	- Semi-structured interviews -Observational Data -Preference assessment -Descriptive evaluation of parent-child interaction	-The participant refrained from speaking to avoid social interaction and SM happened in the presence of multiple strangers. -Treatment increased speech production with unfamiliar people as well as qualitative aspects of speech (responses and initiations). - The parents implemented this treatment in naturalistic settings and the participant exhibited a generalization of the intervention between different people and contexts. -Based on behavioral skills training.	Clinical Case Studies.
Keville et al. (2023)	United Kingdom	Exploring the experiences of parents in caring for children with SM and Autism	Qualitative	11 Mothers and 1 father of autistic children between 5 and 18 years old	Semi-structured interviews	The complex overlap of symptoms resulting from people, environment, and sensory overload impacted the children and their ability to communicate. Judgment and symptom minimization in health and educational settings exacerbated the delay in diagnosis, hindering adequate intervention. The difficulties in caring for a child with SM and Autism, together with broader misunderstandings, increased parental stress and affected the family. Parental advocacy and safe environments facilitated the management of contextual mutism in children. Improvements in detection and compassionate understanding by broader systems, including parents as key stakeholders, are essential for improving the situation.	International Journal of Developmental Disabilities

Ludlow et al. (2022)	United Kingdom	Exploring different factors contributing to social anxiety in children with SM, with and without concurrent Autistic Spectrum Disorder (ASD)	Quantitative	75 Parents	<p>-Selective Mutism Questionnaire (SMQ; Bergman et al. 2008)</p> <p>-Autism Spectrum Screening Questionnaire (ASSQ; Ehlers et al., 1999)</p> <p>- Revised Child Anxiety and Depression Scale, Parent Version (RCADS-P; Chorpita et al., 2000)</p> <p>- Sensory Profile 2 (SP2, Dunn 2014)</p>	<p>-The SM + ASD group exhibited higher levels of social anxiety and sensory avoidance compared to the SM-only group.</p> <p>- Sensory avoidance is a mediator between this diagnosis and social anxiety.</p> <p>- Higher levels of sensory avoidance can help distinguish social anxiety between groups and it can also be a sign of ASD in children with SM who have and/or have not been diagnosed with ASD.</p>	Journal of Autism and Developmental Disorders.
Muris et al. (2021)	London, South Africa, United States.	Examining the relationship between SM and social anxiety, autistic features, and behavioral inhibition to the unfamiliar (i.e., the tendency to respond with restraint and withdrawal when faced with novel stimuli and situations)	Quantitative	<p>Parents of 172 undiagnosed children (96 boys and 76 girls).</p> <p>Aged 3 (n = 45, 26.2%), 4 (n = 61, 35.5%), 5 (n = 42, 24.4%), and 6 (n = 24, 14.0%)</p>	<p>-SMQ (Bergman et al. 2008), Selective Mutism Questionnaire</p> <p>-PAS-R (Edwards et al., 2010), Preschool Anxiety Scale Revised</p> <p>-ASQ (Van der Ploeg and Scholte, 2014), Autism Spectrum Questionnaire.</p> <p>-BIQ-SF (Edwards, 2007), Behavioral Inhibition</p>	<p>- Positive and statistically significant correlations between SM and social anxiety, autistic features, and behavioral inhibition.</p> <p>- Social anxiety and autistic features represented a significant and unique proportion of the variance in SM scores.</p> <p>- These variables stopped contributing significantly once behavioral inhibition was added to the model.</p>	Frontiers in Psychology

					Questionnaire, Short Form		
Muris et al. (2023)	South Africa, Netherlands	To study the psychopathological and temperamental correlations of SM (symptoms)	Quantitative	Mixed sample of non-clinical patients (n=127, 62 girls and 65 boys), and clinically referred patients (n=42, of which 25 showed non- selective syndrome).	The parents completed questionnaires to measure the severity of SM, social anxiety, ASD, and behavioral inhibition of their children.	SM symptoms were related to social anxiety and an anxiety-prone temperament (behavioral inhibition), but they also suggest that ASD-related issues are involved in the selective non-speaking behavior.	Child Psychiatry & Human Development
Steffenburg et al. (2018)	Sweden	To examine the prevalence of ASD in a relatively large clinical sample of children with SM who were referred to a center specialized in neurodevelopmental disorders. A second aim was to analyze possible gender- based differences regarding ASD.	Quantitative	97 Participants (71 girls and 26 boys)	Clinical/diagnostic assessment Cognitive function assessment (Wechsler Preschool or Primary Scale of Intelligence, Wechsler Intelligence Scale for Children, Wechsler Adult Scale of Intelligence). Griffiths' Developmental Scales I and/or II).	-The average age for SM symptom onset was 4.5 years, while the average age for SM diagnosis was 8.8 years. -SM was more frequent among girls (girl : boy ratio = 2.7 : 1). - 63% of the study group had ASD (no difference between genders). - The SM+ASD group showed a later onset of symptoms, older age at diagnosis, more frequent history of speech delay, and a higher proportion of borderline intellectual functioning or intellectual disability.	Neuropsychiatric Disease and Treatment
Valaparla et al. (2018)	India	To present the case of an 11-year-old boy with SM and ASD who was	Case Report, Qualitative	An 11-year- old boy with SM and ASD	Exploration of family history	The effectiveness of non-pharmacological interventions in children with SM and ASD is	Asian Journal of Psychiatry

		effectively treated with non-pharmacological strategies.		and his parents	Mental state examination	highlighted, and it is also established that both conditions can be comorbid.	
Wakamatsu (2022)	Australia	To present a case report involving music therapy intervention for a person experiencing severe SM during the majority of their school years	Qualitative, Case Study	Teresa, 16 years old.	60 music therapy sessions within four years	<p>This case is unique, as most other documented cases involving individuals with SM describe short-term music therapy interventions with very young children shortly after SM has been identified. Teresa's mother had a clear understanding of how Teresa presented with SM; however, given that her formal diagnosis was ASD and ID, and her expressive language abilities could neither be assessed nor observed, her schoolteachers and other therapists had considered her nonverbal rather than selectively mute.</p> <p>Language barriers may also have posed a challenge in advocating for her daughter as she grew older, and she was unable to access effective therapeutic information until Teresa was nearly finished with high school. It is also likely that Teresa's overall cognitive abilities and motor skills were inaccurately assessed, as her performance varied greatly depending on who was observing her.</p>	Australian Journal of Music Therapy.

APPENDIX 2. Study Quality Assessment.

Criterion Score (0-3)	Studies Analyzed							
	Cengher et al. (2021)	Steffenburg et al. (2018)	Muris et al. (2021)	Valaparla et al. (2018)	Ludlow et al. (2022)	Keville et al. (2023)	Muris et al. (2023)	Wakamatsu (2022)
Explicit theoretical framework	3	3	3	1	3	3	3	1
Statement of aims/objectives in main body of report	3	3	3	3	3	3	3	3
Clear description of research setting	3	3	3	1	3	3	3	3
Evidence of sample size considered in terms of analysis	1	3	3	1	3	3	3	1
Representative sample of target group of a reasonable size	1	3	3	1	3	1	3	0
Description of procedure for data collection	3	3	1	1	3	3	3	3
Rationale for choice of data collection tool(s)	3	3	3	3	3	3	3	3
Detailed recruitment data	3	1	1	3	3	3	3	3
Statistical assessment of reliability and validity of measurement tool(s) (Quantitative only)	3	2	3	0	3	0	3	0
Fit between stated research question and method of data collection (Quantitative only)	1	3	3	0	3	0	3	0
Fit between stated research question and format and content of data collection tool e.g. interview schedule (Qualitative only)	1	0	0	1	0	3	0	1
Fit between research question and method of analysis (Quantitative only)	2	3	3	0	3	0	3	0
Good justification for analytic method selected	3	3	3	3	2	3	3	3
Assessment of reliability of analytic process (Qualitative only)	3	0	0	2	0	3	0	1
Evidence of user involvement in design	2	3	3	3	3	3	3	3
Strengths and limitations critically discussed	3	3	3	2	3	3	3	2
Score	38	39	38	25	41	37	42	27
Quality %	79.2%	92.9	90,4%	59.5%	97.6%	88.1%	100%	64.3%