

*EXAMINATION OF FEEDING
DIFFICULTIES AND DYSPHAGIA
IN CHILDREN WITH AND
WITHOUT AUTISM WHO
PRESENT FOR SLEEP
EVALUATION*

Scholarly Pursuit and Thesis
Submission

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Abstract

Research Question:

In children with and without Autism Spectrum Disorder (ASD), how does the presence of various feeding difficulties influence the spectrum of sleep difficulties?

Introduction and Significance:

Autism Spectrum Disorder (ASD) is a complex neurodevelopmental condition characterized by restricted, repetitive patterns of behavior and unusual responses to sensory stimuli. The DSM V has integrated various clinical conditions into the unified diagnosis of ASD, contributing to its diverse nature and variations in age of diagnosis. Sleep and feeding difficulties are noted in the pediatric autism population, however, limited studies explore the relationships between these difficulties. The purpose of this study was to examine the relationships between sleep and feeding disorders in a cohort of children aged 3-17 with and without ASD.

Materials and Methods:

A secondary data analysis was conducted of de-identified data from Nationwide Children's Hospital (NCH) Sleep DataBank, a collection of encounters with children referred for polysomnography. Data were filtered for ages 3-17 years and further filtered for the presence of an autism diagnosis.

Results:

There were 3,053 unique participants ($M = 7.26$ years) with a total of 83,045 encounters between 2006 and 2020. Of these, 239 (8%) were diagnosed with ASD, Asperger's Syndrome, or Pervasive Developmental Disorder. Chi-square analysis revealed a significant association between autism and feeding difficulties, $\chi^2=81.36$, $p<.001$, $OR=3.83$, and dysphagia, $\chi^2=18.95$, $p<.001$, $OR=2.19$. Children with autism were 3.8 and 2.2 times more likely to develop feeding difficulties or dysphagia than children without autism. Since there was a significant association among these variables, feeding difficulties and dysphagia were then used in logistic regressions to predict the likelihood of developing various sleep disorders. In children with autism, those with feeding

difficulties were more likely to develop insomnia (OR=2.12) and sleep disturbance (OR=2.20) than those who are not diagnosed with feeding difficulties. Children with autism who also have dysphagia were more likely to develop sleep apnea (OR=2.30) than children without dysphagia.

Conclusions:

The research reveals a significant correlation, indicating that children with ASD are 3.8 times more likely to experience feeding difficulties and 2.19 times more likely to experience dysphagia compared to neurotypical children. Moreover, the data highlights a link between feeding challenges and sleep disorders, showing that children with ASD and feeding difficulties are more likely to develop sleep disturbances and insomnia. The data also suggests an increased likelihood of sleep apnea in children with ASD who have dysphagia. Overall, these findings emphasize the complex nature of ASD and its co-morbid conditions, underscoring the necessity for an integrated approach in assessing and caring for affected children to improve their overall quality of life. The study advocates for continued research to comprehend the mechanisms underlying these co-morbid conditions and develop effective strategies for early identification and intervention, aiming to enhance clinical practices and support services for individuals impacted by ASD.

Research Question/Aims

This study aims to investigate the relationship between various sleep disorders and feeding difficulties in children with and without Autism Spectrum Disorder (ASD). Sleep disorders encompass a range of issues, from insomnia to sleep apnea, which can significantly impact normal, restful sleep. Feeding difficulties refer to a spectrum of eating patterns that may include limited intake, selectivity, and aversion which can impact a child's overall health and development. Recognizing both sleep disturbances and feeding difficulties can have profound impacts on the developmental trajectory and quality of life of the individual as well as their caregivers. The outcomes of this investigation could offer valuable insights for developing targeted screening guidelines when evaluating children and adolescents in clinical settings.

Specific Aim 1: Investigate the prevalence and types of feeding disorders in children with ASD.

Specific Aim 2: Assess the association between autism and feeding difficulties; and ASD and dysphagia.

Specific Aim 3: Investigate the predictive relationships between feeding difficulties/dysphagia and sleep disorders in children with autism.

Specific Aim 4: Highlight implications for early intervention and treatment

- Discuss the significance of the observed associations between feeding difficulties, dysphagia, and sleep disorders in children with ASD.
- Emphasize the importance of early intervention and treatment for feeding-related issues to improve overall sleep and health outcomes in the pediatric autism population

Introduction

Autism spectrum disorder (ASD) constitutes a subset of neurodevelopmental disorders characterized by challenges in social communication and the presence of restricted, repetitive patterns of behavior, interest, or activities^{1, 2, 3, 4}. The integration of previously distinct clinical conditions, namely autistic disorder, Asperger syndrome, childhood disintegrative disorder, and pervasive developmental disorder not otherwise specified, into the unified diagnosis of ASD, as outlined in the current Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM V).

The prevalence of ASD in children is estimated at 1.7%, although the incidence remains indeterminate due to a lack of consistent longitudinal data in this cohort. The diverse nature of ASD symptoms further complicates the diagnostic landscape, leading to variations in the age of diagnosis. Over time, there has been an observed increase in the estimated prevalence of ASD, attributed to several factors including revisions to the DSM criteria, heightened public awareness promoting increased screening^{2, 3, 5}, and the reclassification of cases that would have historically been categorized as intellectual disabilities or co-occurring genetic syndromes^{2, 6}.

Prevalence rates in the US population align with those of other industrialized countries, while lower rates are reported in resource-limited countries. Regional variations in prevalence may indicate differences in service availability, local provider practices for ASD screening, or community resources⁷. Ethnicity-based disparities exist, as 2014 CDC data revealed that white, non-Hispanic children were approximately 20% more likely to be identified with ASD than non-Hispanic African American children and 50% more likely than Hispanic children².

Prevalence differences also extend to gender in the context of ASD. Comprehensive studies reveal that males are 2-3 times more likely to manifest ASD^{8, 9, 10}. However, females with ASD may be underdiagnosed, as social norms for young females and males vary, influencing the recognition of symptoms consistent with ASD¹¹.

The diagnosis of ASD relies on behaviorally defined clinical symptoms. These symptoms are neurologically based and vary with age, language ability, and cognitive and motor function. They

fall into two primary domains: social communication and restricted repetitive behaviors. The Diagnostic and Statistical Manual of Mental Disorders (DSM-V), outlines specific criteria for ASD, ensuring continuity in diagnosis from the DSM-IV, as nearly all individuals previously diagnosed with autistic disorder or Asperger syndrome would meet the updated ASD criteria under DSM-V.

In addition to the distinctive symptoms linked to ASD, individuals often experience co-occurring conditions. These include seizures, sleep disorders, gastrointestinal (GI) issues, feeding disorders, obesity, catatonia, and developmental conditions such as ADHD, learning disabilities, anxiety disorders, or speech or language disorders². The presence of these conditions influences the health and overall quality of life for children with ASD and places additional stress on their support systems.

The proposed manuscript seeks to bridge a gap in the literature by examining the intersection of feeding difficulties and dysphagia with sleep disturbances within the ASD population. Current research has often explored these challenges as separate entities. However, this paper explores the interdependencies between these co-morbid conditions. This integrated perspective not only advances our understanding of the multifaceted presentations of ASD but also has practical implications for developing targeted interventions that address both feeding difficulties and sleep disturbances, thereby improving the overall quality of life for individuals with ASD and their caregivers.

Methods

A secondary data analysis was conducted of de-identified data from Nationwide Children's Hospital (NCH) Sleep DataBank, a collection of encounters with children referred for polysomnography. Data were filtered for ages 3-17 years and further filtered for the presence of an autism diagnosis. Analysis of the data included reviewing and categorizing ICD-10 codes according to patient encounters, ensuring the exclusion of any repeated ICD-10 codes for individual patients. Analysis of the data included reviewing and categorizing ICD-10 codes according to patient encounters, ensuring the exclusion of any repeated ICD-10 codes for individual patients. Additional demographic information about the de-identified patient information was collected for further analysis.

A chi-square test was conducted to examine the relationship between ASD diagnoses and the occurrence of gastrointestinal (GI), feeding, or sleep disorders. Associations with p-values less than 0.05 were considered statistically significant and were further examined. Within this analysis, the gastrointestinal diagnoses of "feeding disorders" and "dysphagia" were considered, while a separate examination was conducted for the diagnoses of "GERD", "Esophagitis", "Diarrhea", "Gastroenteritis", and "Constipation". Subsequently, a linear regression model was created using only those variables that demonstrated statistically significant p-values in the chi-square test to predict the likelihood of developing various sleep diagnoses if a feeding disorder was present.

Results

The study sample identified 3,053 unique participants across 83,045 encounters who presented for polysomnography evaluation and 239 individuals (8%) were diagnosed with ASD, Asperger's syndrome, or pervasive developmental disorder. Among the participants, 1,709 (56%) were male and 1344 (46%) were female. The racial demographics included 2,015 (66%) white, 620 (20.3%) Black, 229 (7.5%) multiple races, 78 (2.5%) Asian, and 5 (0.1%) Native American, Native Hawaiian or Pacific Islander. A significant majority of 2,864 (93.8%) were non-Hispanic.

The results of a chi-square test indicated a significant correlation between ASD and 12 diagnoses (Table 1). Of these diagnoses, diagnoses outside the scope of feeding difficulties and sleep difficulties were excluded from this analysis and discussed elsewhere. The odds ratios facilitated the assessment of the association's magnitude, wherein the data showed that children with ASD were 3.83 times more likely to develop feeding difficulties and 2.19 times more likely to develop dysphagia than children without ASD.

Variable	Chi square	p value	OR
GER/D	18.806	<.001***	1.93
Esophagitis	7.49	0.006**	3.74
Food hypersensitivity	0.64	0.42	
Lactose Intolerance	0.22	0.64	
Diarrhea	10.98	0.001***	1.75
IBD	1.84	0.18	
NV	5.32	0.02*	1.4
Gastroenteritis	10.56	0.001**	1.9
Abdominal pain	0.96	0.33	
Constipation	42.15	<.001***	2.39
Dysphagia	0.07	0.79	
Celiac	0.27	0.6	
Feeding difficulties	81.36	<.001***	3.83
Dysphagia feeding	18.95	<.001***	2.19
Anorexia	0.22	0.64	
Insomnia	78.79	<.001***	3.51
Parasomnia	2.27	0.13	
Myoclonus	0.22	0.64	
Sleep disturbance	17.63	<.001***	1.77

Excessive daytime sleepiness	0.23	0.63	
Circadian rhythm	11.38	0.001**	1.63
Sleep Apnea	9.6	0.002**	1.55
Daytime sleepiness	10.53	0.001**	1.8
Nocturnal enuresis	1.68	0.19	
Nocturnal leg movements	5.93	0.02*	1.6
Tired	4.02	0.045*	1.62
Snoring	0.52	0.47	
Inadequate sleep hygiene	0.33	0.56	
*= $p < .05$, **= $p < .01$, ***= $p < .001$			

Table 1. Chi-square results examining the association between being diagnosed with autism vs no autism and having a concurrent GI, feeding, or sleep diagnosis.

In the regression model exhibited in Table 2, only those variables that demonstrated statistically significant p-values in the chi-square test were incorporated. As the aim of this paper specifically pertains to disorders of feeding, feeding diagnoses included in the model were “feeding difficulties” and “dysphagia”. These predictor variables were used in the models to predict the likelihood of developing various sleep diagnoses if a feeding disorder was present. The sleep diagnoses that were included (dependent variables) were insomnia, sleep disturbance, circadian rhythm, sleep apnea, daytime sleepiness, nocturnal leg movements, tired, and snoring.

Sleep Disorder	Predictor	B*	p**	Exp(B)***
Insomnia				
	Diarrhea	0.989	0.014	2.69
	NV	0.82	0.018	2.269
	Constipation	0.749	0.023	2.115
	Feeding Difficulties	0.786	0.018	2.195
Sleep Disturbance				
	Feeding Difficulties	0.79	0.024	2.203
Circadian rhythm				
	NV	0.832	0.015	2.297
Sleep Apnea				
	Dysphagia Feeding	1.115	0.014	3.05
Daytime sleepiness				
	NV	0.806	0.056	2.239
Tired				

	Constipation	1.125	0.043	3.08
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Table 2. Results of binary logistic regressions for patients **with** ASD.

*The regression weight value, denoted B, quantifies the impact of each predictor variable in the binary regression.

**The p-value, denoted p, indicates the variables of significance. Variables with a p-value less than 0.05 were noted to be significant.

***The odds ratio, denoted Exp(B), is the predicted change in odds for a unit increase in the predictor.

Additionally, children with ASD and feeding difficulties had a higher likelihood of experiencing insomnia (OR 2.12) and sleep disturbances (OR 2.20) compared to those without feeding difficulties.

Furthermore, children with ASD who had dysphagia were more likely to develop sleep apnea, with a ratio (R) of 2.30 compared to those without dysphagia. These findings underscore the intricate interplay between feeding challenges and sleep disorders in children with ASD.

Discussion

The results of this study indicated a markedly higher possibility of developing feeding difficulties and dysphagia in children with ASD as compared to children without ASD (Figure 1), 3.8 times and 2.19 times, respectively. This suggests a strong likelihood there is a linkage between ASD and the diagnoses of both “feeding difficulties” and “dysphagia”. This relationship has previously been explored as previous studies estimated approximately three out of four children with ASD experience eating-related problems. Feeding difficulties often manifest as food selectivity influenced by sensory aspects of food such as color, texture, temperature, and food presentation rituals ^{7, 12}. This selectivity stems from behavior rigidity, social impairments, and sensory processing difficulties, leading to issues like pica and rumination, or the act of self-stimulated emesis and swallowing of stomach contents ¹³.

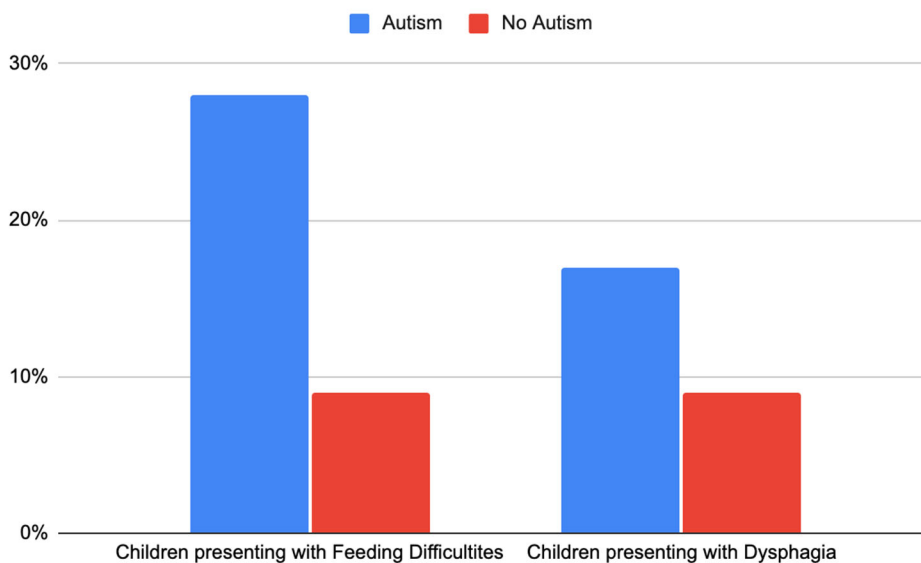


Figure 1. Percentage of children with and without ASD presenting with feeding difficulties and dysphagia

In the literature concerning children with ASD, feeding difficulties related to food selectivity, have been called selective eating, food fussiness, picky eating, food refusal, food aversion, and atypical eating ⁷. Sensory hyper-responsiveness can make mealtimes overwhelming, triggering avoidance, while hypo-responsiveness may result in disinterest in eating ^{13,14}. Additionally, children with ASD may have delayed oral motor skills, affecting their ability to manage certain textures. Of note, while children with ASD often receive an adequate volume of nutrition, resulting in anticipated

weight gain, their diets often lack variety ¹⁵. Therefore, there is a heightened risk of nutritional deficiencies due to food aversions, especially to fruits and vegetables ^{16,17,18}, which can significantly impact a child's health, potentially leading to conditions like scurvy ^{19, 20, 21}, rickets, keratoconus ²², and calcium deficiency ^{13, 23, 24}.

Furthermore, the elevated incidence of insomnia and sleep disturbances in children with ASD who have feeding difficulties points to a connection between feeding challenges and sleep patterns. The relationship between ASD and sleep disturbances is thought to be multifactorial involving environmental, biological, psychological, and co-occurring health conditions. Previous research has indicated sleep disorders impact 50-80% of individuals diagnosed with ASD ^{25, 26}. In a study involving 1518 children with ASD aged between 4 and 10 years, it was found that 71% of children experienced sleep disturbances, despite only 30% being formally diagnosed with a sleep disorder ²⁷. The odds ratio (OR) of 2.12 for insomnia and 2.20 for sleep disturbance found in this analysis of children with ASD and feeding difficulties indicates a greater than two-fold increase in risk in the development of either sleep condition.

Dysphagia, a condition characterized by difficulty swallowing, represents any interruption in the coordinated sequence of muscular contractions and relaxations that facilitate swallowing. It can compromise not only the safety and efficacy of swallowing but also the adequacy of an individual's nutritional status ²⁸. There are many etiologies of dysphagia and if left untreated, can lead to aspiration, malnutrition, dehydration, respiratory infections, and an overall diminished quality of life. Although not as widely reported as feeding difficulties, dysphagia is thought to be common in children with ASD, with the resultant complications mirroring those found in the neurotypical population ^{29, 30}. The observation found in this analysis that children with ASD and dysphagia are more likely to develop sleep apnea (R = 2.30) is of note and could suggest a physiologic link between swallowing mechanisms and sleep-disordered breathing. More studies are needed to elucidate the mechanism.

This study builds upon existing research regarding the occurrence of feeding difficulties and dysphagia in children with ASD, contributes novel analysis regarding the interaction between feeding difficulties and sleep disturbances and insomnia in children with ASD as well as the

interaction of dysphagia and sleep apnea in children with ASD. The findings suggest the need for integrated care approaches that address both feeding and sleep concerns in children with ASD. The findings of this study suggest early identification and intervention regarding feeding difficulties could reduce the risk of developing associated sleep disorders. However, as this study was constructed around observational data, causality cannot be proven between the variables. Given the proposed overlap in symptoms suggested in these findings, it underscores the importance of a multidisciplinary approach when caring for children with ASD.

Future Directions

The results from the retrospective case-controlled study conducted using the dataset published by Nationwide Children's Hospital identified a significant relationship between Autism Spectrum Disorder (ASD) and the co-occurrence of feeding difficulties and certain sleep disorders. Given these results, there is a need for research into targeted therapeutic interventions such as behavioral therapies, dietary modifications, and sleep hygiene tailored to children with ASD. Such interventions could improve the overall quality of life for both the child as well as the child's caregivers.

Longitudinal studies are needed to examine the developmental patterns of both feeding and sleep disorders in children with ASD. These analyses could aid in determining if early feeding difficulties are predictive of the onset of sleep disorders, or if the relationship is bidirectional in that early sleep disorders are predictive of feeding difficulties.

Additionally, although the dataset utilized to conduct this analysis was large, it consisted of a higher percentage of white, non-Hispanic participants. Future studies should aim to include a more diverse population to ensure the findings are representative and generalizable to a broader population. Including a more ethnically and socioeconomically diverse population would also allow for the analysis of such factors' influence on the management of ASD, feeding difficulties, and sleep disorders.

The relationship between ASD and feeding difficulties as well as ASD and dysphagia, necessitates further research regarding targeted therapeutics specific to this population. These interventions could include behavioral therapies, dietary modifications, and sleep hygiene practices fitted to children with ASD. Such interventions could not only improve the health of the child but also improve the quality of life of the child's caregiver.

Lastly, research on the physical and psychological effects of the complex caregiving needs of children with ASD and co-occurring conditions is crucial. Insights from such research could guide interventions to reduce the resultant caregiver stress and improve relationships between the child and caregivers.

Conclusion

This study examines the relationships between ASD, feeding difficulties, dysphagia, and sleep disturbances. The findings demonstrate a significant association between ASD and increased risks of developing both feeding difficulties and dysphagia wherein children with ASD are 3.8 times more likely to experience feeding difficulties, and 2.19 times more likely to experience dysphagia, compared to neurotypical children. This analysis also illustrated the relationship between feeding challenges and sleep disorders, with children with ASD and feeding difficulties being more likely to develop sleep disturbances and insomnia. Additionally, the data suggests that in children with ASD, those with dysphagia were more likely to develop sleep apnea than those without dysphagia.

These findings expand upon previous literature regarding the multifactorial nature of ASD and its co-morbid conditions. The intersection of feeding difficulties and dysphagia with the sleep conditions described throughout this analysis underscores the need for an integrated approach when assessing and caring for children with ASD to enhance the overall quality of life for individuals with ASD and their support systems.

As these comorbid conditions are likely to remain a persistent problem for children with ASD, future research should aim to further understand the mechanism underlying the relationship of these co-morbid conditions and to develop effective strategies for early identification and intervention. This will contribute to the body of knowledge about ASD while improving clinical practices and support services for those impacted directly and indirectly by ASD.

Compliance

The analysis has been granted exemption from Institutional Review Board (IRB) oversight due to the de-identification of the data and its initial collection for non-research purposes, rendering it publicly available. This determination ensures compliance with ethical standards while recognizing the minimal risk associated with the use of de-identified data collected for non-research purposes.

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