

Food selectivity in autism spectrum disorders: A systematic review

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ABSTRACT

Autism spectrum disorders (ASDs) are characterized by difficulties with reciprocal social interactions, deficiency in verbal and non-verbal communication, and restricted, repetitive and stereotyped patterns of behavior and interest; one of these characteristic behaviors is food selectivity. The objective of this study was to perform a systematic

review of the literature published between 1970 and 2013 concerning this eating behavior. The articles identified were analyzed in terms of: sample size, study design, and criteria for assessment and intervention, as well as the results, level of evidence and grade of recommendation.

The main search was conducted in Medline, although other databases were also consulted (Cochrane Library, Scielo, ScienceDirect and Embase). Regarding the methodological quality of the articles retrieved, almost all had limited statistical validity for some of the following reasons: lack of a control group and/or definitions of inclusion criteria, analysis being based on single individuals, heterogeneous groups including different ages, small sample sizes, or failure to consider phenotypic variability between individuals or alternative explanations, as well as multiple meanings being applied to the term "food selectivity". Despite all this, there is empirical evidence and an overall scientific consensus supporting an association between food selectivity and ASDs.

Key words: feeding assessment, selective eating, nutrition, growth, BMI, autism, ASD (Autism Spectrum Disorder), feeding disorders, mealtime problems, review.

INTRODUCTION

Autism spectrum disorders (ASDs) are characterized by an impaired ability to engage in social interactions as well as a deficit of verbal and non-verbal communication, and a restricted, repetitive and stereotyped patterns of behavior and interests (Filipek, 1999). These include "food selectivity", though it is important to recognize that in the literature this term has multiple meanings, being used to refer to a range of concepts including food refusal, a limited repertoire of accepted foods and high-frequency single food intake. Indeed, the lack of consensus complicates the assessment and comparison of results from different studies. What does seem clear is that the problems related to food intake are varied, from mild cases in which the behaviors do not pose a risk to health, to more severe cases in which there is a risk of malnutrition or exceeding recommended intakes, and some requiring tube feeding (Williams et al, 2010). On the other hand, ASDs are associated with depression or violent behaviors that are frequently the consequence of attempts by individuals with these disorders to control their environment. Other common and non-specific characteristics include anxiety and sleeping disorders. Lastly, gastrointestinal (GI) disorders have also been described.

ASDs are syndromes that affect psychological and cognitive development, with an estimated prevalence of 5 cases per 10,000 worldwide according to the criteria for classical autism of Kanner and of 6.6 cases per 1000 considering all the spectrum (CDC, 2007; Neuschaffer, 2007), although these figures are based on data from 2002. On the basis of more recently published data for 2008 (CDC, 2012), it is estimated that the prevalence has increased to 11.3 cases per 1000 people (1 in 88 people), being

more common in males (ratio of 4:1). No statistically significant differences have been detected as a function of socioeconomic level or between the various cultures studied (Bagdahli, 2005).

Behavior and physiological perspective: Behavioral problems play an important role in food habits of children with ASDs. Their eating patterns tend to be governed by food aversion/refusal or preferences for certain types of food at the expense of others. Some of the factors involved include the texture, color, taste, shape and temperature of food, as well as shape and color of the packaging or layout and presentation of dishes, and even the types of utensils used. In some cases, however, it is possible to identify physiological factors that are the direct or indirect cause of certain behavioral and eating problems including impaired sensory processing or difficulties with oral motor skills such as chewing and swallowing, and GI disorders.

Both delayed development and more severe impairment of fine and gross skills are more common in children with ASDs than controls (Provost et al., 2007; Ming et al., 2007; and Matson et al., 2011). In addition, Brisson et al. (2012) reported more problems related to motor anticipation in children with ASDs than among typically developing (TD) children. That is, children with ASDs may not have the necessary motor skills for handling food and the resulting behavioral response may be fear, aggression or escape. It is important that interactions between the sensory and motor systems are not ignored since an exclusively behavioral approach to treating these children could underrate the impact of oral sensory problems on their feeding habits. Indeed, in children with “food selectivity” accompanied by nausea, vomiting and/or choking the problems may well be secondary to sensory disorders and, in such cases, food refusal can be considered an adaptive behavioral response (Overland, 2011). Tachtman-Reilly et al. (2008) note that ASD children have problems related to sensory modulation (hypo- and hyper-reactive) in hearing, sight, smell, taste and touch, as well as with vestibular system and proprioception. On the other hand, when there are no identifiable organic factors (abnormal sensory processing, oral motor disorders, or GI problems), food selectivity can be considered to be the manifestation of the restricted interests and the behavioral rigidity characteristic of ASDs (Ledford and Gast, 2006).

Taking a behavioral perspective, Hsu and Ho (2009) observed a higher rate of eating rituals in ASD than TD children. Overall, the etiology of the eating problems seems to involve physiological weaknesses together with sensory, motor, cognitive and emotional dysfunction (Lockner et al., 2008), repetitive behaviors and restricted interests (core characteristics of autism), being among the consequences of these other problems. Martins et al. (2008) and Nadon et al. (2011) also described this phenomenon.

With regards to medical problems, many authors (Dohan, 1966; Goodwin et al., 1971; Cade et al., 2000; Erickson et al., 2005; Xia et al., 2010; Adams et al., 2011; Wang et al., 2011; and Souza et al., 2012; among others) have observed higher rates of GI disorders in ASD than TD children. On the other hand, Black et al. (2002) and Whitehouse et al. (2011) did not find these differences and, similarly, Ibrahim et al. (2009), after grouping the symptoms that characterize GI disorders (constipation, diarrhea, abdominal distension, gastroesophageal reflux and food selectivity) only found statistically significant differences for constipation and food selectivity, both

attributable to the neurobehavioral disorder, ASD, rather than a primary digestive problem. Maenner et al. (2012) demonstrated that certain behaviors associated with ASDs, such as food refusal, were significantly associated with GI disorders, although these behaviors were common regardless of the presence of GI problems, suggesting that these associations may be of limited diagnostic value.

Another, no less important issue, is chewing difficulties due to dental problems. There is relatively little in the literature on oral health in people with ASDs, in particular, the prevalence of caries, and the results are not conclusive. Among the studies that have focused on this issue, Namal et al. (2007) and Subramaniam et al. (2011) reported a lower prevalence of caries in control TD children, while Loo et al. (2008) and Jaber (2011) found the reverse, and De Moor et al. (1997) found no significant differences between ASD and TD children,

Prevalence: Food selectivity is not exclusive to children with ASDs or other disabilities. Approximately a quarter of all children have eating problems during their first years of life, though the rate may be as high as 80% in children with developmental disabilities (Bryant-Waugh et al, 2010). It should be underlined that, though this does not disappear completely, it tends to improve with age. Nadon et al. (2011) found that nearly 90% of preschool and school-age children with ASDs do not process sensory information, in particular related to touch, smell, sight and hearing, in the same way as their TD peers. On the other hand, Kerwin et al. (2005) stated that although more than 60% of parents interviewed reported that their children with ASDs had strong food preferences, only 6.7% of them described their children as not having an appetite.

Cornish (1998) found that 70% of ASD children in their study sample were selective eaters. More recent studies have confirmed this pattern. For example, Williams et al. (2000) indicated that overall 67% of parents complained of food selectivity, it being determined by the following factors: texture (69%), appearance (58%), taste (45%), smell (36%), and temperature (22%), as well as reluctance to try new foods (69%) and a small repertoire of accepted foods (60%). Similarly, Schreck et al. (2006) found that 72% of parents reported their children having a limited repertoire of foods and 57% reported food refusal. Foods were mainly rejected due to their presentation, the use of certain utensils or different types of food being served on the same plate (48.6%). Other factors related to refusal/acceptance included: the characteristics of the tableware (13.8%) and texture of the food (6.5%), as well as oral motor disorders (23.2%). Consistent with this, a study focused on assessing the eating habits and intake of nutrients in children with ASDs, by Schmitt et al. (2008), found that 70% of children with ASDs selected what they ate according to texture compared to 11% of children without ASD. Whiteley et al. (2000) indicated an even higher rate (83%) of parents reporting that their children ate a restricted repertoire of foods, while Klein and Nowak (1999) found that just over half (53%) of their patients were unwilling to try new foods. In general, Bandini et al. (2010) indicated that children with ASDs rejected food more often than TD children (41.7% of foods offered compared to 18.9%).

Given all of the above, we considered that it would be useful to conduct a review to assess the state of knowledge on food selectivity. The resulting description of studies reported to date, in terms of the number of participants and other characteristics, is intended to serve as a summary of the available scientific evidence, both for the

research community and for professionals involved in diagnosis, assessment and implementation of interventions in people with ASDs.

METHODS

We conducted a systematic review of the scientific literature from 1970 to 2013 related to food selectivity. As a guide for evaluating the articles, we characterized the level of evidence they provided, according to the methodological quality in terms of study design, using these levels as a basis to grade the associated recommendations. Specifically, the identification of information sources was based on using the Medline Database, in accordance with the recommendations of the Spanish National Health System, though occasionally other databases were also consulted (Cochrane Library, Scielo, ScienceDirect and Embase). In the search, the following key words were used: feeding assessment, selective eating, nutrition, growth, BMI, autism, ASD, autism spectrum disorders, feeding disorders, mealtime problems, and review, with the corresponding Boolean operators. The last search was performed on 11 January 2013.

For classifying the level of evidence and grade of recommendations for the articles reviewed, we used the instrument proposed by the European Association for the Study of Obesity (Tsigos C et al., 2008), which is a simplified version of the system developed by the Scottish Intercollegiate Guidelines Network (2008). The scale proposes that two characteristics of the source be used for assessing the quality of the scientific evidence provided (level of evidence): the study design and the risk of bias. Numbers from 1 to 4 are used to rate the study design, while signs (++ , + and -) indicate the assessed risk of bias, according to the degree of fulfillment of key criteria related to this potential risk. Based on this assessment of the quality of the scientific evidence in the source, grades are used to classify the strength of associated recommendations

In addition to the aforementioned system of levels, we considered the following features, as applicable, to assess the level of evidence provided by the selected articles: (A) Degree of homogeneity of the group studied (as determined by definitions and criteria applied); (B) use of a control group and the appropriateness of the selection; (C) type of experimental design (randomized or not); (D) knowledge of the intervention by patients, relatives and other observers (open, simple or double blind trial); (E) nature of the dietary regimen (level of strictness) and degree of adherence; (F) selection of assessment criteria, including the instruments used (questionnaires, scales, etc.) for assessing changes in patient status under the treatment; and (G) the presence of confounding factors including any types of pharmacological treatments provided, or the use of one or more intervention procedures that could affect the assessment criteria selected.

For evaluating and synthesizing the scientific evidence, we also considered the internal validity of the studies, whether there was statistical significance and the accuracy of the results (confidence intervals and effect size), as well as their clinical relevance. We then characterized the recommendations on the basis of the quantity, generality, and clinical relevance of the results as well as the quality of the scientific evidence. Overall, the studies were analyzed by consideration of the following: sample size, study design and nature of the intervention (in such cases), assessment criteria, and the biomedical

and behavioral findings themselves, as well as the level of evidence and grade of recommendation.

This paper complies with the methodological requirements established for the publication of systematic reviews (Friedenreich CM, 1993; Froom P et al, 1993) and the PRISMA recommendations (Liberati A et al, 2009; Urrutia G, 2010). We initially considered all the systematic reviews and full original articles published from 1970 to date that were retrieved, as well as the abstracts of the references reported in those articles. Publications were then selected for analysis if they met the following criteria: the participants, of any age, met the DSM-IV-TR criteria for ASDs; and the dependent variables were related to potential changes in biomedical or behavioral symptoms observed in patients with food selectivity (e.g., increase in intake or food acceptance). Finally, we did not limit the search by language but studies that did not assess final health outcomes were excluded.

RESULTS

In the articles reviewed, in addition to reporting improvements in eating behaviors, mention is made of changes in anthropometric parameters as well as improvements in nutritional status. The earliest analysis of eating behaviors in children with ASDs pointed out that they accept or reject foods according to their nature (O'Bannion et al., 1978). Subsequent studies by Ahearn et al. (2001) and Field et al. (2003) confirmed this selectivity, with half of affected children being selective eaters. On the other hand, neither of these studies used a control group and hence their validity is very weak. The lack of a control group is a serious issue, considering that children with normal psychomotor development may be picky and fussy about food without their nutritional status being affected (Davis, 1928).

Schreck et al. (2004, 2006) and Tomchek et al. (2007) found heightened smell and taste sensitivities in children with ASDs. Twachtman-Reilly et al. (2008), comparing ASD and TD children, demonstrated that those with ASDs have a more limited repertoire of accepted foods, a trend that was confirmed by Sharp et al. (2010), and Seiverling et al. (2010, 2011) reported a preference for soft and semi-liquid foods. Over the years, many other studies have referred to dietary selectivity in children with ASDs (Raiten et Massaro, 1986; Clark et al., 1993; Ahearn et al., 2001; Ahearn, 2003; Najdowski et al., 2003; Buckley et al., 2005; Luiselli et al., 2005; Dominick et al., 2007; Keen, 2008; Martins et al., 2008; Schmitt et al., 2008; Johnson et al., 2008; Cermak et al., 2010; Emond, 2010; Maskey et al., 2012; and Zimmer et al., 2012). Overall, the available evidence suggests that this selectivity may imply a higher risk of nutritional deficiencies that could, in turn, affect anthropometric characteristics, considering that height and weight curves, as well as the body mass index are, although indirect, a priori ideal methods for assessing nutritional status (AF, 1983; Gordon CC, 1988). Table I summarizes sample sizes, study designs, assessment criteria and interventions, and results of the studies analyzed, as well as the levels of evidence and grades of recommendation.

DISCUSSION

Conceptual constraints: In the articles retrieved, "food selectivity" carries a variety of meanings from food refusal, to having a restricted repertoire of foods or the frequent

intake of a single type of food. It is also used to refer to the classification of foods as a function of their nutritional components (rich in proteins or in fats) or sensory characteristics (e.g., textures such as smooth, soft or hard, and tastes such as sweet or salty). There are no standard criteria for defining severities of “intake problems”, these ranging widely from mild conditions with no likely health risks, to more severe cases given the potential for malnutrition or exceeding recommended intakes, and extreme cases in which tube feeding is required.

The use of terms with multiple meanings is also a problem regarding definitions of GI symptoms (constipation, diarrhea, abdominal distension, gastroesophageal reflux, and food selectivity). Lastly, there is still a lack of standardized definitions of the various ASDs themselves, those available so far being limited or incomplete, and it is to be hoped that this may be rectified with the changes proposed in the new versions of the diagnostic criteria, DSM-V due to be published later this year. All this makes it difficult to compare the results between studies and is a potential source of bias.

Methodological constraints: Almost all the articles found had validity issues for various reasons: lack of a control group, analysis being based on single individuals or anecdotal data, groups that were heterogeneous in term of age, small sample sizes, or no statement of inclusion criteria, as well as a failure to consider phenotypic variation between individuals or alternative explanations. There is another potential source of bias in the behavioral variables, namely that in the reports of parents or caregivers memories may be distorted over time and may include subjective perceptions, for example, regarding the behavior of participants, associated with the mere fact of participating in a clinical trial.

Limitations of the review process: Our search may not have retrieved all the relevant publications on the subject and the review itself may be sensitive to information bias.

Recommendations: Overall, in the trials of behavioral interventions, significantly greater improvements were observed in terms of calorie intake per meal and weight gain in groups with ASDs than in controls, when available, or reference populations (matched by age and sex). Given this, we conclude that behavior at meal times should be monitored as part of the routine follow-up of patients with ASDs. In addition, sensory-motor skills should be assessed systematically, as should the presence of possible GI disorders. On the other hand, considering that all the articles reviewed were unanimous that food selectivity is associated with ASDs, pediatricians as well as parents and other caregivers of people with autism, especially those with dietary restrictions, must in any case be aware of the potential seriousness of deficiencies. Indeed, the prevention of deficiencies should be a high-priority nutritional objective, encouraging children with ASDs to eat a balanced, varied, moderate, sufficient and pleasant diet. Nevertheless, further research is required including, in particular, blinded randomized, controlled trials with larger sample sizes.

AUTHOR CONTRIBUTIONS

All five co-authors of this paper (SM, IZ, AM, MM and AL) have contributed significantly to the design and implementation of the study, as well as the analysis and interpretation of the results. Further, all have participated in the preparation of this manuscript and have approved the final version submitted for publication.

CONFLICTS OF INTEREST

No authors have any conflicts of interest to declare.

Table I. Studies on food selectivity in ASDs

AUTHOR GR	N	DESIGN	ASSESSMENT CRITERIA	RESULTS	LE
O'Banion D et al. (1978)		One 8-year-old child with ASDs.			
		No control group and others worsened it	Case study 3	Behavioral D	Certain foods improved behavior
Raiten DJ, Massaro T		(1986)	Cases: 40 children with ASDs		
		Controls: 34 TD children	Observational,		
		Case-control study	7-day food diary, Parental questionnaire	Significantly more food selectivity in children with ASDs	2+ C
Cornish E		(1998)	17 children with ASDs between 42-117 months old.		
		No controls	Observational,	Cross-sectional	Parental interview,
		FFQs,			
		3-day food diary	Reported food preferences,		
		Problems due to texture, color and presentation.	3	D	
Whiteley et al. (2000)		100 children with ASDs 2-16 years old.			
		No controls.	Observational,	Cross-sectional	Parental reports
		Reported food preferences and eating difficulties	3	D	
Williams PG et al. (2000)		100 children with ASDs 1.8-10 years old			
		No controls	Observational,	Cross-sectional	Parental interview
		Reported food preferences and eating difficulties	2-		
Ahearn WH et al. (2001)		30 children with ASDs 3-14 years old.			No
		control group	Observational, Open-label,		
		Cross sectional	Refusal/dietary preferences	50 % showed refusal	3 D
Lesinskiene S et al. (2002)		Cases: 25 children with ASDs, mean: 11 years old.			
		Controls: 50 TD children, mean: 6.5 years old.	Open-label,	Case-control	
		Parental questionnaire	Lower level of appetite and smaller range of food consumed	2-	

Ahearn WH Case study D	(2003)	One child with ASD 14 years old Dietary modification, Behavioral	Experimental, Improvement in intake.	3
Najdowski AC Case study D	(2003)	One child with ASD 5 years old Dietary modification, Behavioral	Experimental, Decrease in food selectivity	3
Field D et al.	(2003)	349 children (26 with ASDs) between 1-144 months old Open-label, retrospective.	Selectivity-predisposing factors GERD was the most important factor for refusal (50%)	3 D
Schreck KA et al.	(2004)	Cases: 138 children with ASDs, Controls: 298 DT children.		
Both between 5-12 years old, participation 25%/30%. Case-control		Food acceptance patterns questionnaire	Observational, Open-label, More food selectivity and eating difficulties in children with ASDs	2-
Buckley SD et al. Case study D	(2005)	One 9-year-old girl with ASD Dietary modification, Behavioral	Experimental. Improvement in food selectivity	Case study 3
Luiselli JK et al. Case study	(2005)	One 4-year-old girl with ASD Gradual exposure to milk	Experimental,	
Behavioral assessment 3 D			Improvement in food selectivity: acceptance of milk	
Schreck KA et al.	(2006)	138 children with ASDs 5-8 years old. Open-label, observational, cross-sectional.	No controls. Food acceptance patterns questionnaire More food selectivity in children with ASDs than their relatives	3 D

The studies classified as 1– or 2– were not used in the recommendation process given the high risk of bias.

Abbreviations: ASD: Autism Spectrum Disorder; TD: typical developing; FFQ: food frequency questionnaire. GERD: Gastroesophageal reflux disease; EAR : Estimated Average Requirement; LE: Level of evidence; and GR: Grade of recommendation

Table I. Studies on food selectivity in ASDs (cont.)

AUTHOR	N	DESIGN	ASSESSMENT CRITERIA	RESULTS	LE GR
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Dominick KC et al. (2007) Cases: 67 children with ASDs,

Controls: 39 children with language impairment,

All 4-14 years old Case-control. Parental interview, Standardized questionnaires
Significantly more food selectivity in children with ASDs 2-

Tomchek SD (2007)

Cases: 281 children with ASDs, Controls: 281 TD children,

All 3-10 years old Open-label,

Age matching, Case-control. Sensory processing More smell and taste sensitivity in children with ASDs 2+ C

Lockner DW et al. (2008) Cases: 20 years with ASDs,

Controls: 20 TD children, age and sex-matched,

All 3-5 years old. Observational. Case-control. Parental reports Poorer perception of dietary behaviors among parents with children with ASDs compared to those of DT children

2+ C

Keen DV (2008) 7 children with ASDs Descriptive, Case series
Relationship between ASD and anthropometric measurements Feeding problems and lower growth 3 D

Schmitt L et al. (2008) Cases: 20 children with ASDs,

Controls: 18 TD children,

All 7-10 years old Observational. Case-control. FFQ,

3-day food diary Greater difficulty due to texture and less variety in children with ASDs 2-

Bandini LG (2008) Cases: 53 children with ASDs,

Controls: 58 TD children,

All 3-11 years old Observational. Open-label. Case-control. FFQ.
3-day food diary, Parental interview More food selectivity in children with ASDs 2-

Martins Y et al. (2008) Cases: 58 children with ASDs,

Controls: 137 TD children,

All 2-12 years old Observational. Open-label. Case-control.
Feeding and eating behavior questionnaire to mothers More food selectivity and eating difficulties in children with ASDs 2-

Williams KE et al. (2010) Systematic review The behavioral intervention significantly improved food selectivity and prior treatment of medical conditions such as GERD is important 2-

Emond A (2010) Cases: 79 children with ASDs,

Controls: 12901 TD children,

All between 6-54 months old Observational, case-control. FFQ, anthropometric measurements "Difficult to feed", "very demanding and selective" 2+ C

The studies classified as 1– or 2– were not used in the recommendation process given the high risk of bias.

Abbreviations: ASD: Autism Spectrum Disorder; TD: typical developing; FFQ: food frequency questionnaire. GERD: Gastroesophageal reflux disease; EAR: Estimated Average Requirement; LE: Level of evidence; and GR: Grade of recommendation

Table I. Studies on food selectivity in ASDs (cont.)

AUTHOR	N	DESIGN	ASSESSMENT CRITERIA	RESULTS	LE	GR
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Maskey M et al. (2012)		863 patients with ASDs, 2-18 years old.				
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No controls	Review of case studies	Parental questionnaire: Assessment of behavioral and emotional problems				
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	53% had eating behavior problems		3			D
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Sharp WG et al. (2012)		Systematic review		The behavioral intervention significantly improved food selectivity	2++	B
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Zimmer MH et al. (2012)		Cases: 22 children with ASDs,				
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		Controls: 22 TD children,				
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		all 8-year-olds Observational,				
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		age-matched, case-control.		Nutritional adequacy, FFQ,		
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		.EAR. Significantly more food selectivity in children with ASDs	2+			C
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The studies classified as 1– or 2– were not used in the recommendation process given the high risk of bias.

Abbreviations: ASD: Autism Spectrum Disorder; TD: typical developing; FFQ: food frequency questionnaire. GERD: Gastroesophageal reflux disease; EAR : Estimated Average Requirement; LE: Level of evidence; and GR: Grade of recommendation.

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