Prevalence of Stereotypy in Individuals with Developmental Disabilities: A Systematic Review

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Abstract

Although many researchers have examined the prevalence of stereotypy in individuals with developmental disabilities, the results of previous studies have not been aggregated and analyzed methodically. Thus, we conducted a systematic review of studies reporting the prevalence of stereotypy in individuals with developmental disabilities. Our results indicated that the average prevalence of stereotypy across studies was 61% and that individuals with autism spectrum disorders had the highest reported prevalence (i.e., 88%) across specific diagnoses. Children and adults generally had similar overall prevalence measures, but the specific forms varied with age and diagnosis. Studies using the Repetitive Behavior Scale – Revised and the Autism Diagnostic Schedule – Revised generally reported higher estimates of prevalence of specific forms of stereotypy when compared to the Behavior Problem Inventory. However, the latter seemed more sensitive than the Aberrant Behavior Checklist for overall prevalence. Studies with a low risk of bias found a lower prevalence of stereotypy than those with a high risk of bias. Our results underline the importance of continuing research efforts to improve the assessment and treatment of stereotypy in individuals with developmental disabilities.

Keywords: autism spectrum disorder, developmental disability, prevalence, repetitive behavior, stereotypy

Prevalence of Stereotypy in Individuals with Developmental Disabilities: A Systematic Review

Autism spectrum disorders are characterized by impairments in social communication as well as by the presence of unusual, repetitive and restricted behaviors and interests, which interfere with daily functioning (American Psychiatric Association [APA], 2013). As such, many individuals with ASD engage in repetitive motor and vocal behaviors, commonly referred to as stereotypy in the research literature (Cunningham & Schreibman, 2008; Lanovaz & Sladeczek, 2011). These behaviors are also common in individuals with other types of developmental disabilities (Lundqvist, 2011, 2013). Researchers typically define stereotypy as involuntary, patterned, repetitive, coordinated, rhythmic, and non-reflexive behaviors that are not mediated by social stimuli (Freeman, Soltanif, & Baer, 2010; Rapp & Vollmer, 2005). Stereotypy may take on several forms such as body rocking, mouthing, repetitive hand movements, finger flicking, spinning, twirling, mouthing objects, toe walking, pacing, hand waving, object banging, repetitive vocalizations, and repetitive posturing (Bodfish, Symons, Parker, & Lewis, 2000; MacDonald et al., 2007).

Even though typically developing children often engage in stereotypy at a young age, repetitive behaviors tend to decrease past two years old (Berkson & Tupa, 2000; MacDonald et al., 2007; Thelen, 1979). Concerns therefore arise when stereotypy (a) persists at similar levels past the age of two, (b) is displayed with high intensity or frequency, (c) appears atypical or unusual in its manifestation, or (d) interferes with an individual's functioning (Didden et al., 2012). From clinical and educational standpoints, the problem with stereotypy is that engaging in the behavior may interfere with the acquisition of new skills (Dunlap, Dyer, & Koegel, 1983; Lanovaz, Robertson, Soerono, & Watkins, 2013; Matson, Kiely, & Bamburg, 1997; Morrison &

Rosales-Ruis, 1997) and with social interactions (Reese, Richman, Belmont, Morse 2005) in addition to being socially stigmatizing (Jones, Wint, & Ellis, 1990).

Given its adverse repercussions on development and integration, examining the prevalence of stereotypy in different populations with developmental disabilities appears important. To this end, multiple research teams have reported the prevalence of stereotypy within these populations (e.g., Bhattacharyya, Sanyal, Roy, & Saha, 2009; Bishop et al., 2013; Bodfish et al., 1995; Cuccaro et al., 2007; Goldman et al., 2009; Lundqvist, 2011, 2013; McTiernan, Leader, Healy, & Mannion, 2011). For example, Goldman et al. (2009) found that 69% of children with ASD engaged in at least one form of stereotypy whereas McTiernan et al. (2011) obtained an overall prevalence of 92% within the same population. Similarly, Bhattacharyya et al. (2009) reported that 63% of individuals with Down syndrome engaged in at least one form of stereotypy; in contrast, Lundqvist (2013) found a prevalence of 31%. Given that the results differ from one study to another, we believe that it is important to compare the results obtained by different research teams and to examine the impact of variables that may potentially explain these discrepancies (e.g., participant characteristics, measurement method, selection procedures).

To our knowledge, no study has systematically aggregated and analyzed the prevalence of stereotypy in individuals with developmental disabilities across studies. Although some studies have compared prevalence across diagnoses and to a more limited extent age (e.g., Flores et al., 2011; Lundqvist, 2011; Woodcock, Oliver, & Humphreys, 2009), these results were not compared with other studies and researchers have not conducted an analysis of further important variables such as scales used and potential sampling bias. Thus, our study aims to systematically review previous research pertaining to the prevalence and forms of stereotypy displayed by

individuals with developmental disabilities. Addressing this issue could better inform prevention and intervention efforts and help direct resources where they are needed most.

Method

Search Procedures

A systematic literature search of articles was performed in July 2014 and again in May 2015 using the PubMed and PsycInfo electronic databases. We designed search terms to include population with autism spectrum disorders and other developmental disabilities (autism or autistic or asperger or pervasive development* or intellect* disabilit* or down* syndrome or trisomy 21 or mental* retard* or developmental disorder* or developmental disabilit* or rett syndrome or developmental delay or delayed development), stereotypy or description of behaviors associated with stereotypy (stereotyp* behavior* or stereotypy or self stimulation or sensory reinforcement or self reinforcement or automatic reinforcement or aberrant behavior* or repetitive vocalization* or repetitive behavior* or ritualistic behavior* or self-stimulatory behavior* or repetitive motor mannerism or vocal stimulation or hand flapping or echolalia or eye gazing or body rocking or toe walking or hand clapping or object twirling or finger wiggling or pacing or mouthing or tapping or jumping or sniffing) and names of scales commonly used to evaluate stereotypy or related behaviors (observation* method* or aberrant behavior checklist or stereotyped behavior scale or repetitive behavior scale or behavior problems inventory or stereotypy severity scale or stereotypy linear analog scale or nisonger child behavior rating form or diagnostic assessment for the severely handicapped* or repetitive behavior questionnaire). We limited our search for articles published in 1994 and later, the year of publication of DSM-IV by the APA.

In total, the PubMed search yielded 266 records and the PsycInfo search yielded 611. The removal of duplicates left a total 721 records to be screened. The first two authors independently screened the titles and abstracts of the articles identified. We retrieved full-text articles if they were primary studies of individuals with developmental disabilities of any age and if they were reporting information about stereotypy or if further clarification regarding the fulfillment of inclusion criteria was required. A hand search of the reference lists of the included articles yielded 16 additional articles.

The same authors independently read the full texts of 478 articles and included studies if they were (a) in English; (b) peer-reviewed; (c) included participants with an intellectual disability, an ASD, a genetic syndrome, or a combination thereof; (d) provided information on the proportion of study participants that presented stereotypy or on the proportion of participants that showed a specific form of stereotypy; and (e) were accessible. Moreover, they excluded studies that (a) presented the results of behavioral or pharmacological interventions; (b) only reported an average score on a stereotypy scale or only showed the information on a graph as opposed to reporting a specific proportion; (c) reported stereotypy and other behavior (e.g., tics, compulsions, self-injury) in the same undifferentiated category; and (d) presented only an aggregated prevalence of individuals with and without a developmental disability. If agreement on inclusion could not be reached, the third author was consulted. In total, 44 studies met our inclusion and exclusion criteria, with 11,743 participants included in the review. Figure 1 illustrates the results of the search and of the screening and selection process for the inclusion of studies in our review.

Data Collection and Analyses

For each included study, the first author extracted the prevalence measures. Articles could include an overall prevalence of stereotypy, the prevalence of specifics forms, or both. The overall prevalence was defined as the proportion of individuals in the sample that engaged in at least one form of stereotypy (i.e., regardless of form) at a given point in time. This measure was only reported when the study presented an overall value including multiple forms of stereotypy. If the study only reported specific forms (but no overall value), we did not report an overall prevalence for that specific study. Given that the specific forms and categories of stereotypy varied considerably from one study to another, we classified forms of stereotypy according to seven categories: 1. Whole body (e.g., rocking, swaying), 2. Head (e.g., head rolling, head nodding), 3. Hand/Finger (e.g., hand flapping, clapping, finger flicking and wiggling), 4. Locomotion (whirling, jumping, bouncing), 5. Object usage (e.g., twirling, banging, throwing) 6. Sensory (e.g., gazing, mouthing, smelling) and 7. Vocal (e.g., echolalia, repetitive nonsensical sounds). The first six categories were based on the stereotyped behavior subscale of the Repetitive Behavior Scale – Revised (RBS-R; Bodfish et al., 2000). We added the seventh category, vocal stereotypy, as research suggests that it may be relatively frequent in children with ASD (MacDonald et al. 2007).

Table 1 presents a list of examples of forms that we included in each category. When researchers presented the prevalence of multiple forms from the same category within an article, we reported the highest prevalence as our purpose was to estimate the percentage of individuals who engaged in at least one form of stereotypy within a category. Similarly, if a study presented the results of more than one scale, we used the highest value in our analyses. Lastly, when a study presented a prevalence of multiple forms from two or more categories together (e.g., head

and body stereotypy), we did not include the value in our analysis as we were unable to categorize it.

Next, we aggregated the results of the studies together and compared the prevalence of stereotypy across diagnoses, age groups, scales used, and risk of bias. To aggregate the results for each analysis, we computed the median prevalence of stereotypy as an average. We chose to use the median rather than the weighted mean because we wanted to avoid studies with a high risk of bias and a large number of participants exerting more weight on the average than the ones with a low risk of bias, therefore skewing the results. If we had used weighted means, well-designed studies with a low number of participants would have exerted a negligible weight on any final value given some studies had more than 1,000 participants. It is also worth mentioning that we only calculated and presented a median for a given analysis when at least three studies reported a prevalence measure. We used descriptive rather inferential statistics to present our results as the number of studies per category were often too low and the samples too diverse to have sufficient power to conduct further statistical testing.

For our initial analysis, the first author categorized the articles by diagnoses. When an article only included participants from one specific diagnostic category (e.g., ASD, Fragile X, Down syndrome), we reported the prevalence under the label of the diagnosis. When the article presented an aggregated value of prevalence which included multiple diagnostic categories or only individuals with intellectual disability with unspecified or unknown etiologies, we reported the value under the label “developmental disability”. Lastly, if the article presented specific values for multiple diagnoses, we reported the values for each specific diagnosis as well as an aggregated value of all participants under the label developmental disability.

For our age analysis, we divided the studies into two groups: children and adults. If a study only included individuals between the ages of 0 and 17 years, we included it in the children's category. Alternatively, if a study only included individuals 22 years of age or older, we included it in the adults' category. Because the bounds for studies of children and adults often included ages between 18 and 21, we applied the following additional guideline. If the maximum age was between 18 and 21 years old and most of the sample's participants were less than 18 years old, we included the study in the children's category. If the minimum age was between 18 and 21 years old and most of the sample's participants were older than 21, we included the study in the adults' category. Studies that included both children and adults (i.e., did not meet any of the above criteria) were not included in this analysis.

In our review, we identified five scales that were frequently used to report the prevalence of stereotypy: the Diagnostic Assessment for the Severely Handicapped (DASH-II; Matson, Smiroldo, & Hastings, 1998), the Aberrant Behavior Checklist (ABC; Aman, Singh, Stewart, & Field, 1985), the Behavior Problem Inventory (BPI; Rojahn, Matson, Lott, Esbensen, & Smalls, 2001), the RBS-R (Bodfish et al., 2000), and the Autism Diagnostic Interview- Revised (ADI-R; Lord, Rutter, Couteur, 1994). Thus, we also compared stereotypy prevalence based on the scale used. With the exception of the ADI-R, all scales were informant-based questionnaires. The DASH-II includes subscales for assessing the presence of anxiety, self-injurious behavior, elimination and eating disorders, mood, sleep and sexual disorders, organic syndromes, impulse control problems, schizophrenia, pervasive developmental disorder, and the presence of stereotypic behaviors. The ABC consists of five subscales assessing: a) irritability, agitation and crying, b) lethargy and social withdrawal, c) stereotypic behavior, d) hyperactivity and non compliance, and e) inappropriate speech. The BPI is comprised of a self-injurious behavior

subscale, a stereotyped behavior subscale and an aggressive/destructive behavior subscale whereas the RBS-R assesses the presence of ritualistic behavior, stereotypic behavior, self-injurious behavior, compulsive behavior and restricted interests. Unlike the other tools, the ADI-R is a structured interview conducted with parents of children suspected of having ASD. The interview covers a wild range of behaviors in the area of social interaction, communication, and repetitive behaviors.

To examine the potential effect of selection bias on prevalence estimates, we additionally assessed whether the selection of study participants could have influenced the reported results. A study was judged as having a low risk of bias if a consecutive or a random sample of participants in the population with the targeted diagnosis was enrolled or if the study avoided inappropriate exclusions; an unknown risk of bias if no information was provided on the selection of participants or a high risk of bias if a purposive or a convenience sample of participants was enrolled (Whiting et al., 2011). We hypothesized that studies having a high risk of bias in participants' selections (lack of random sampling, targeting of participants because of their increased risk of displaying stereotypy) would produce higher prevalence estimates of stereotypy than studies with a low selection bias. Two authors gathered and verified data from the 44 articles.

Results

Table 2 presents the prevalence of stereotypy reported across studies by alphabetical order (based on the first author's last name) while Table 3 presents the medians for the overall prevalence of stereotypy and the prevalence of specific categories of stereotypy based on the participants' diagnoses, age groups, scales used, and risk of selection bias. When taking into account all studies, the median overall prevalence of stereotypy is 61% in individuals with

developmental disabilities. In general, studies reported similar prevalence of hand/finger (*Mdn* = 49%), locomotion (*Mdn* = 46%), object (*Mdn* = 54%), sensory (*Mdn* = 47%) and vocal (*Mdn* = 48%) stereotypy whereas the least prevalent forms were whole body (*Mdn* = 30%), and head stereotypy (*Mdn* = 26%). As for prevalence of stereotypy for specific diagnoses, individuals with ASD displayed stereotypy the most frequently with an 88% average, followed by individuals with Fragile X syndrome (*Mdn* = 69%), Down syndrome (*Mdn* = 63%), and Prader-Willi syndrome (*Mdn* = 44%). An insufficient number of studies (i.e., < 3) reported overall prevalence for other diagnoses (e.g., Cornelia de Lange, Williams syndrome), preventing us from computing a median. Individuals with ASD engaged the most frequently in sensory stereotypy (*Mdn* = 73%) and the least often in head stereotypy (*Mdn* = 30%).

We also compared the prevalence of stereotypy of adults to that of children. Interestingly, the average prevalence of stereotypy of adults (*Mdn* = 61%) was similar to children (*Mdn* = 57%). Our results also indicate that children engaged the most frequently in sensory stereotypy (*Mdn* = 70%) and the least often in head stereotypy (*Mdn* = 26%). The adults on the other hand, engaged the most frequently in whole body stereotypy (*Mdn* = 50%), and similarly to children, engaged the least often in head stereotypy (*Mdn* = 29%). As for the scales used, the BPI generally produced higher estimates of overall prevalence (*Mdn* = 85%) than the ABC (*Mdn* = 33%). For the prevalence of specific forms, the ADI-R yielded similar estimates to the RBS-R. In contrast, the BPI consistently produced lower estimates than the RBS-R for specific forms.

Lastly, we examined whether prevalence of stereotypy varied depending on the study's risk of bias in the selection of participants. Studies suffering from a high risk of bias found that on average, 84% of individuals with developmental disabilities engaged in stereotypy as opposed to studies with a low risk of selection bias that produced an average overall prevalence estimate

of 58%. Studies with an unknown risk of bias whereby the authors did not provide us with enough information regarding participants' selection to make an informed decision regarding their selection bias reported that on average 61% of individuals with developmental disabilities engaged in stereotypy.

Discussion

Taken together, our results indicate that most individuals with developmental disabilities engage in at least one form of stereotypy. Moreover, the diagnostic category associated with the highest overall prevalence was ASD. Children and adults with developmental disabilities had similar prevalence estimates. The RBS-R and ADI-R generally produced higher estimates of prevalence of specific forms of stereotypy when compared to the BPI, but the latter seemed more sensitive than the ABC for overall prevalence. Not surprisingly, studies with a low risk of bias found a lower prevalence of stereotypy than those with a high risk of bias. Furthermore, our results indicate that the most and least prevalent forms of stereotypy varied depending on the participants' diagnoses, age groups and scales used.

Our results extend research by aggregating the findings of previous studies and examining the impact of variables such as age group, diagnosis, and scale used on prevalence estimates. The specific disability reported the most often was ASD, which was expected given that previous studies have highlighted the contribution of autism severity and IQ in the prevalence of stereotypy (Goldman et al. 2009; Matson, Wilkins, & Macken, 2008). Conversely, syndromes such as Cri du Chat, Cornelia de Lange, Williams, and Angelman were not the subject of sufficient studies to provide estimates. As for the scales used to measure prevalence of stereotypy, the BPI, the ABC, the RBS-R, and the ADI-R were used the most often. Our analyses suggest that the RBS-R may be more sensitive to specific forms and the BPI to overall

prevalence. Based on our results, clinicians should expect most children and adults with whom they work to engage in at least one form of stereotypy. The least prevalent form regardless of age group was head stereotypy, but whole body was most prevalent in adults and sensory in children, which may be an artefact of the sample distributions (i.e., the ASD diagnosis is overrepresented in studies of children when compared to adults). These results further support the importance of continuing research on the assessment and treatment of specific forms of stereotypy in individuals with developmental disabilities, which may improve functioning while reducing stigmatization.

The use of multiple scales, various age ranges, and differing diagnostic categories across studies may limit the results of our systematic review. The items evaluating specific forms within a category of stereotypy differed across studies; as such, the forms included within each category varied based on the scale used. Most studies reported stereotypy for specific forms (rather than for a stereotypy category); in these cases, we reported the most prevalent form. Our medians are most likely lower bounds of the actual prevalence in the population given that most forms within a category were not usually mutually exclusive. Similarly, some scales excluded certain forms of stereotypy. For example, the stereotyped behavior subscale of the RBS-R does not include a specific item for vocal stereotypy and the BPI only includes one item (i.e., yelling and screaming), which most likely led to an underestimation of both overall and specific prevalence.

The lumping of various diagnoses together in some studies adds to the complexity of the results and to their representativeness to each syndrome alone. It should also be noted that the changes in criteria in the definitions of both ASD and intellectual disabilities in the latest version of the DSM may also have an impact on current prevalence (APA, 2013). When more studies are conducted using the DSM-V for the diagnosis inclusion criteria, our study should be replicated to

compare whether the prevalence and forms of stereotypy remain the same. The comparison process is limited by the recruitment methodology that differed from one study to the next; thus, the representativeness of the results often remained unknown. Our research strategy also had its limitations that should be noted. We searched only two databases and we relied on the indexation of the articles to retrieve relevant studies. Hand searches allowed us to retrieve more articles, but some may have been missed. In addition, we did not contact authors for studies where no information was provided on the selection of participants in order to assess the risk of bias; we rated them as unknown.

Despite these limitations, our analyses yield several recommendations for future research on the prevalence of stereotypy. First, researchers should continue to directly compare different diagnostic groups together using the same scale and to report the prevalence of each group separately. Few studies have done so (see Flores et al., 2011 and Lundqvist, 2011, 2013 for notable exceptions), which is why adopting a comparative approach could be highly relevant in the future. Researchers should also conduct more prevalence studies on diagnoses that were under-represented in our systematic review (e.g., Cornelia de Lange, Cri du Chat, Williams syndrome). To provide more complete and precise estimates of prevalence, we recommend adding more items specific to vocal stereotypy within widely used scales (e.g., RBS-R, BPI). Future research should also aim to reduce bias in participant selection as most of the studies reviewed had a high risk of selection bias. As noted earlier, studies with a lower risk of bias tended to produce lower estimates of prevalence. Taking into account those issues could potentially improve the validity and reliability of prevalence estimates in the research literature. In turn, having access to better prevalence estimates may eventually improve service delivery by guiding practitioners and researchers in their intervention and research efforts.

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Table 1

Examples of forms included in each stereotypy category

Whole body	Head	Hand/Finger	Locomotion	Object	Sensory	Vocal
Rocking	Head movements	Hand, finger stereotypy	Whirling, turning around	Object usage	Gazing	Yelling
Body stereotypy	Head nodding	Hand, finger movements	Pacing	Watching same video continuously	Sensory stereotypy	Screaming
Clenching-stiffening-posturing	Head rolling	Hand, finger mannerisms	Jumping	Twirling things	Rubbing self	Repetitive talk about one topic
Whole body stereotypy	Head stereotypy	Hand flapping or shaking	Bouncing	Spinning objects	Repetitive behavior involving sensation	Echolalia
Body and torso tensing movements		Finger wiggling	Running	Repetitive use of objects	Sniffing objects	Stereotyped and repetitive use of language
		Waving or shaking arms	Locomotion	Manipulating objects	Eye/vison	Verbal rituals
			Twirling (on one's self)	Object stereotypy	Ear/hearing	Facial grimacing and vocalization
					Mouthing	Vocal stereotypy
					Unusual sensory interest	

Table 2

Prevalence of Overall and Specific Forms of Stereotypy Across Studies

					Proportion of individuals engaging in at least one form of stereotypy (%)								
Study	Scale	Age	Diagnosis	N	Overall	Whole body	Head	Hand/ finger	Loco-motion	Object	Sensory	Vocal	Risk of bias
Baumgardner et al. (1995)	ABC	3-18 *M* = 9	DD	61	21	-	-	-	-	-	-	-	High
			Fragile X	31	11	-	-	-	-	-	-	-	
Bhattacharyya et al. (2009)	DASH-II	3-39 *M* =16	DD	140	61	-	-	-	-	-	-	-	Unknown
			DS	70	63	-	-	-	-	-	-	-	
Bishop et al. (2013)	RBS-R	4-18 *M* =9	ASD	1825	-	26	22	56	53	46	73	-	Unknown
	ADI-R	4-18 *M* =9	ASD	1825	-	-	-	51	-	59	68	-	Unknown
Bodfish et al.(1995)	Other	18-62 *M* =34	DD	210	61	81	43	54	39	63	64	26	Low
Capone et al.(2005)	Other	3-13 *M* =9	ASD	87	-	-	-	-	-	-	-	59	Low
Chadwick et al. (2005)	Other	11-17 *M* =13	DD	82	33	-	-	-	-	-	-	-	Low
Chowdhury et al. (2010)	ADI-R & RBS-R	19-28 *M* =23	ASD	34	-	-	-	50	-	30	44	62	High
Clarke & Marston (2000)	ABC	5-33 *M* =11	Angelman syndrome	73	-	-	-	-	-	-	49	-	High
Cox et al. (1999)	ADI-R	1-3 *M* =21	ASD	21	-	-	-	38	-	33	5	0	Low

					Proportion of individuals engaging in at least one form of stereotypy (%)								
Study	Scale	Age	Diagnosis	N	Overall	Whole body	Head	Hand/ finger	Loco-motion	Object	Sensory	Vocal	Risk of bias
Cuccaro et al. (2007)	RBS-R	4-22 *M* =11	ASD	46	-	35	26	61	54	15	72	-	High
Cuccaro et al. (2012)	ADI-R	4-21 *M* =9	ASD	577	-	-	-	-	-	77	-	-	High
Davies et al. (1998)	Other	19-39 *M* =27	Williams syndrome	70	86	-	-	-	-	-	-	-	High
Dykens & Clarke (1997)	ABC	2-40 *M* =12	Cri du chat syndrome	146	52	-	-	-	-	-	-	-	High
Fecteau et al. (2003)	ADI-R	7-20 *M* =13	ASD	28	-	-	-	62	-	71	78	48	Unknown
Flores et al.(2011)	RBS-R	2-18 *M* = 10	DD	252	-	33	27	62	53	52	70	-	Low
		M = 10	ASD	207	-	37	30	68	63	60	77	-	
		M=11	PWS	45	-	13	13	36	9	16	36	-	
Fostad et al.(2012)	Other	1-3 *M* =2	DD	624	-	33	-	-	-	22	-	22	Low
Goldman et al.(2009)	Other	2-8 *M* =5	DD	277	38	-	-	-	-	-	-	-	High
		M = 5	ASD	129	69	-	-	-	-	-	-	-	
Greaves et al. (2006)	Other	3-18 *M* =11	DD	169	84	-	-	-	-	61	-	-	High
		M =11	PWS	80	79	-	-	-	-	50	-	-	
		M =10	ASD	89	88	-	-	-	-	71	-		

					Proportion of individuals engaging in at least one form of stereotypy (%)								
Study	Scale	Age	Diagnosis	N	Overall	Whole body	Head	Hand/ finger	Loco-motion	Object	Sensory	Vocal	Risk of bias
Hattier et al. (2013)	BPI	1-3 *M* =2	DD	25	-	24	16	56	56	36	32	32	Unknown
		M =2	ASD	13	-	39	31	62	69	54	39	31	
Hessl et al. (2008)	BPI	8-24 *M* =16	Fragile X	50	98	-	-	50	-	-	-	-	Low
Hill & Furniss (2006)	DASH-II	8-29 *M* =18	DD	82	79	-	-	-	-	-	-	-	High
		M =19	ID w/o ASD	13	39	-	-	-	-	-	-	-	
		M =18	ID with ASD	69	86	-	-	-	-	-	-	-	
Hiraiwa et al. (2007)	Other	2-31	PWS	165	-	-	-	-	-	-	-	59	High
Huxley et al. (2005)	ABC	37-63 *M* =49	DS	34	100	-	-	-	-	-	-	-	High
Kim & Lord (2010)	Other	0-4 *M* =2	DD	456	85	-	-	-	-	-	-	-	High
			ASD	336	97	-	-	-	-	-	-	-	
Lam et al. (2007)	RBS-R	3-48 *M* =15	ASD	307	-	46	38	74	53	55	80	-	High
Lam et al. (2008)	ADI-R	1-29 *M* =9	ASD	316	-	25	-	47	-	64	-	-	Unknown

					Proportion of individuals engaging in at least one form of stereotypy (%)								
Study	Scale	Age	Diagnosis	N	Overall	Whole body	Head	Hand/ finger	Loco-motion	Object	Sensory	Vocal	Risk of bias
Lundqvist (2011, 2013)	BPI	18-87 *M* = 43	DD	915	41	11	4	7	8	-	5	14	Low
			PWS	9	44	-	-	-	-	-	-	-	
			DS	113	31	-	-	-	-	-	-	-	
			Fragile X	14	71	-	-	-	-	-	-	-	
			ASD	143	73	-	-	-	-	-	-	-	
McTiernan et al. (2011)	BPI	3-14 *M* =8	ASD	174	92	-	-	-	-	-	-	-	Unknown
Medeiros et al.(2014)	BPI	17-60 *M* =30	DD	115	81	-	-	-	-	-	-	-	Low
Murphy et al.(2009)	BPI	3-14 *M* =9	ASD	157	27	-	-	-	-	-	-	-	Unknown
Myrbakk et al. (2008)	ABC	14-72 *M* =40	DD	140	33	-	-	-	-	-	-	-	Low
Newman et al. (2015)	BPI	2-17 *M* =8	Fragile X	47	100	-	-	-	-	-	-	-	High
Oliver et al. (2009)	ABC	4-22 *M* =14	DD	100	-	45	63	92	-	-	-	-	High
		M =14	CdLS	53	-	62	87	96	-	-	-	-	
Richler et al. (2007)	ADI-R	1-9	DD	214	-	-	-	46	-	70	70	-	Low
			ASD	165	-	-	-	54	-	79	78	-	
Rojahn et al.(1997)	Other	27-49 *M* =37	DD	560	-	50	29	44	36	30	39	57	High

					Proportion of individuals engaging in at least one form of stereotypy (%)								
Study	Scale	Age	Diagnosis	N	Overall	Whole body	Head	Hand/ finger	Loco-motion	Object	Sensory	Vocal	Risk of bias
Rojahn et al. (2001)	BPI	14-91	DD	432	54	18	4	15	11	-	-	22	Low
Rojahn et al. (2012)	BPI	2-93 *M* =34	DD	1122	85	26	-	19	-	15	21	40	High
Rojahn et al. (2013)	BPI	1-61 *M* =17	CdLS	180	87	26	14	37	37	23	45	48	High
Scahill et al. (2014)	Other	4-17 *M* =8	DD	291	-	-	-	15	-	28	16	49	Unknown
Szatmari et al. (2006)	ADI-R	2-13 *M* =8	ASD	339	-	21	-	57	-	61	69	-	High
South et al.(2005)	Other	8-20 *M* =14	ASD	39	-	45	-	48	60	65	-	82	High
Woodcock et al. (2009)	RBQ	6-19 *M* =14	DD	79	33	-	-	-	-	-	-	-	High
		M =14	PWS	46	28	-	-	-	-	-	-	91	
		M =14	Fragile X	33	39	-	-	-	-	-	-	73	
Zeilinger et al.(2011)	ABC	18-80 *M* =40	DD	270	1	-	-	-	-	-	-	-	Unknown

Note. CdLS refers to Cornelia de Lange syndrome; DS: Down syndrome; ID: Intellectual disability; ASD: Autism spectrum disorder; PWS: Prader-Willi syndrome; ABC: Aberrant Behavior Checklist, ADI-R: Autism Diagnostic Interview Revised; RBS-R: Repetitive Behavior Scale – Revised; BPI: Behavior Problem Inventory; RBQ: Repetitive Behavior Questionnaire; DASH-II: Diagnostic Assessment for the Severely Handicapped-II.

Table 3

Median prevalence of stereotypy across diagnoses, age groups, scales used and risk of bias

	Median							
	Overall	Whole body	Head	Hand/ finger	Locomotion	Object	Sensory	Vocal
Diagnosis								
DD (all studies)	61	30	26	49	46	54	47	48
ASD	88	36	30	57	57	61	73	54
Fragile X	69	-	-	-	-	-	-	-
PWS	44	-	-	-	-	-	-	-
DS	63							
Age group								
Children	57	33	26	56	54	60	70	48
Adults	61	50	29	47	36	30	42	42
Scale								
ABC	33	-	-	-	-	-	-	-
ADI-R	-	-	-	49	-	64	69	-
RBS-R	-	34	-	62	53	49	73	-
BPI	85	25	9	28	24	23	21	32
Risk of bias								
High	84	40	34	50	53	43	47	59
Low	58	33	27	46	39	53	39	22
Unknown	61	24	-	56	-	59	53	48

Note. ASD: Autism spectrum disorders; PWS: Prader-Willi syndrome; DS: Down syndrome; ABC: Aberrant Behavior Checklist; ADI-R: Autism Diagnostic Interview Revised; RBS-R: Repetitive Behavior Scale – Revised; BPI: Behavior Problem Inventory

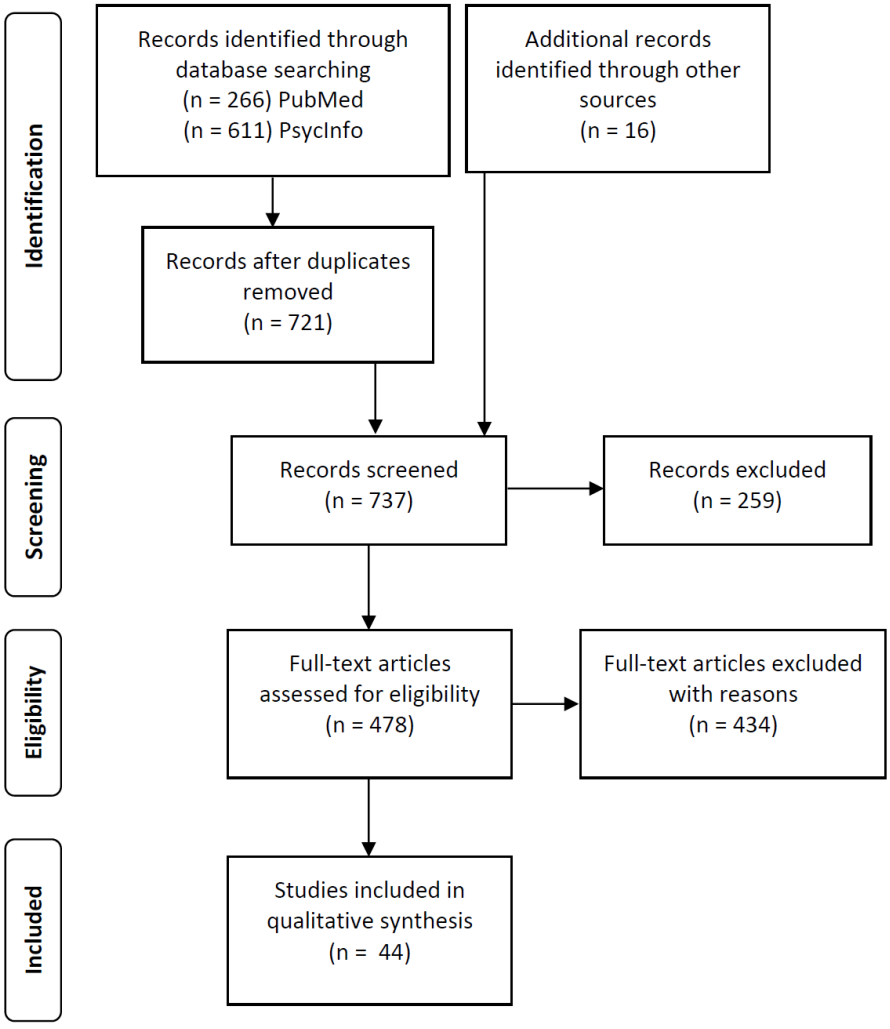

Figure 1. PRISMA flow diagram (Moher et al., 2009)