Clinical Approach

Iran J Pediatr
Jun 2010; Vol 20 (No 2), Pp:149-159

Clinical Approach to Motor Stereotypies in Autistic Children

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Received: *Dec 29, 2009*; Final Revision: *Mar 10, 2010*; Accepted: *Mar 15, 2010*

Abstract

This is an overview of stereotypic behavior in autistic spectrum disorder (ASD). This repetitive, nonfunctional, fixed pattern of behavior is associated with autism severity but it is not specific for ASD. There are a wide range of behaviors mentioned as stereotypies. It usually starts in early childhood and its severity is associated with outcomes and severity of autism in adolescence and adulthood. It is usually co-morbid with other psychiatric problems and its pathophysiology is not exactly known. Management is most likely behavioral. There are some reports regarding efficacy of antipsychotics for its management. Further studies should be conducted to improve our knowledge about it and our ability to differentiate it from tics.

Iranian Journal of Pediatrics, Volume 20 (Number 2), June 2010, Pages: 149-159

Key Words: Motor; Stereotypy; Autism; Clinical Approach; Children

Introduction

Restricted repetitive behaviors (RRB) and stereotypic behaviors (SB) count among the key symptoms of autism. Movement disorders such as stereotypies indicate the severity and progression rate of Rett disorder[1], and the severity of autism symptoms and pragmatic competence at later ages[2]. Social involvement of children with autism with their peers increases their adaptive behavior skills and improve outcome of the disorder[3]. Repetitive and stereotyped movements with objects in children with autism spectrum disorders late in the second year of life predict unique variance in the severity of autism symptoms in the fourth year beyond that predicted by social communication measures alone[4]. So, intervention program on cognitive abilities should be focused and started in early ages so that its influence continues into adolescence and adulthood[3].

More studies are required to be conducted to improve knowledge about the pathophysiology of stereotypies in autism[5]. This is a review for definition, classification, epidemiology, and management of motor stereotypies in children with autism.

Definition of Motor Stereotypies

Motor stereotypies are suppressible, repetitive, rhythmical, coordinated, purposeless, fixed, and

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Fig1: Motor stereotypies

nonfunctional pattern of movements (Fig 1)[6,7].

These movements may happen together and many times in day[7]. The periodic movements are high-frequency. However, rhythmicity is not a characteristic of stereotypy. Stereotypies in autism are associated with severity of autism[8] and lower cognitive development[9]. However, another study did not find association of autism severity and motor stereotypies[6]. The repertoire and manner of movement for each individual is specific. More than one type of stereotypies is usually seen at one moment.

Excitement[10], stimulation, stress, anxiety, boredom, fatigue, sensory isolation, or social demands increase stereotypies[6,7]. Different types of stereotypic behaviors are displayed in Table 1. Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) defines stereotypies as a repetitive and non functional behavior lasting 4 or more weeks. It also emphasizes that the behavior interferes with normal activity or it may lead to self injury.

Classification

The rate of repetitive behaviors in PDD is higher than those with mental retardation[11]. Some authors classified repetitive behaviors into two distinctive groups of 'lower-order' and 'higher-order' sub-groups. The lower-order repetitive behaviors are associated more with developmental delays while the higher-order behaviors are correlated with autism[11]. Another classification classifies stereotypies into two groups of 1) primary or physiological; this type does not have any specific cause for stereotypies such as pencil tapping, hair twisting, and 2) secondary or associated with other conditions such as neurological, sensory problems, with pervasive developmental disorder (PDD), tumor, or infection. For example gazing atypically at objects may be present in PDD[7].

Underlying Disorders

The continuum of repetitive behaviors can be seen in typically developing children[10,12] and it is not limited to autism spectrum disorders (ASD) (Table 2)[6,13].

It also can be seen in children with developmental delay or sensory deprivation[7] syndromes such as Smith-Magenis Syndrome[14] and Cri-du-Chat syndrome[15]. The number and diversity of stereotypies in autism is more than in typically developing children[6]. The rare behavior of atypical gazing at fingers and objects was only observed in autism[6]. Some authors report that self-injurious behavior is a more rigorous type of stereotyped movements and self-injurious behavior is rarely performed in the absence of other stereotyped movements[16].

Autism spectrum disorders

Autism is one of the most common types of autism spectrum disorders. These disorders are behavioral syndromes with various degrees of social impairments, verbal, nonverbal and as well as restricted or stereotyped interests and

Table 1: Different types of stereotypic behaviors

Type	Definition
Face	Grimacing, lips or tongue movements, opening the mouth, mouth stretching, licking[17], smoking, puffing noise[18], sucking objects[16].
Head and neck	Head tilting, shaking, nodding, hair twirling, head banging, neck stretching[19], teeth grinding[20], hair pulling[20], tongue wagging, biting bottoms[18], neck extension.
Trunk	Body rocking, spin[12], spinning or rotation of entire body[17].
Shoulders	Bending, scrunching;arching the back;shrugging the shoulders.
Arm/leg	Arm flapping, bilateral repetitive movements involving the arms and hands such as crossing the arms on the chest, stamping the feet, tapping one's feet, heel and toe walking[17].
Hand/finger	Hand flapping, slapping, nail biting[21], finger wiggling, Shaking, tapping, waving, clapping, opening-closing, rotating or twirling the hand or fingers, thumb-sucking, pointing, fanning fingers[17], fluttering fingers in front of the face, picking skin, scratch self, arrange objects[16].
Hand/finger with object	Shaking, tapping, banging, twirling an object, tapping pencils, touching, rubbing, repetitive ordering[22], arrange toys in patterns[12], adding objects to a line[17], manipulating of objects[16].
Gait	Pacing, jumping, running, skipping, spinning.
Self-directed	Covering the ears, mouthing, smelling, rubbing the eyes, tapping the chin, slapping self or an object or surface, touching genitals, self-mutilating behavior[22].
Visual	Atypical visual explanatory behaviors[23] such as staring at an object or the fingers 'out of the corner of the eyes, eyelid closure, squinting eyes[17].
Vocal and speech	Vocalization, humming, tongue clucking, echolalic words/phrases[24], telling or asking[22].

activities. The age of onset is before 3 years. The other types of autism spectrum disorders are Aspeger's syndrome, Rett's disorder, childhood disintegrative disorder, and Pervasive Developmental Disorder, Not Otherwise Specified (PDD-NOS). Etiology of these disorders is not clearly known[25].

Autism spectrum disorders impact different aspects of the children and also their families, parents, and siblings[26]. The rate of symptoms of ASD in community is considerable[27].

Medications such as antipsychotics and serotonin specific reuptake inhibitors are suggested for management of autism spectrum disorders[28].

Epidemiological factors

The mothers reported point prevalence of stereotypic movement among 3079 children of 1-15 years age in the primary health care centers referred for vaccination was 2.3%[29]. It usually starts before age 3 years[7,30]. Boys more than girls are afflicted and its ratio is about 3:2[7,10,31,32]. Stereotypic behavior levels in 2-, 3- and 4-year-old children with autism or PDD-NOS is more than in the typically developing same-age peers[17]. Even, infants with autism show stereotypic behavior[33]. Forty-four percent of children with autism have at least one subtype of stereotypy[6].

Table 2: Some of the most common underlying diagnoses for stereotypies

Typically developing children
Autism spectrum disorders
Some developmental conditions
Neurological problems

Developmental age is not associated with the presence of repetitive behaviors in autism but lower chronological age is associated more with simple or low-level repetitive behaviors[34].

Stereotypies are more common in children with autism than cognitively-matched non-autistic developmentally disordered children. The occurrence, number, and variety of stereotypies are higher in autism co-occuring with mental retardation (nonverbal Intelligence Quation (IQ)[6].

Key points:

- One of the key features of autism spectrum disorders is restricted repetitive behaviors (RRB) and stereotypic behaviors.
- Motor stereotypies are suppressible, repetitive, rhythmical, coordinated, purposeless, fixed, and nonfunctional pattern of movements.
- Motor stereotypies usually start before age 3 years.
- Stereotypies can be assessed using Repetitive Behavior Scale-Revised (RBS-R) questionnaire or Repetitive and Restricted Behaviour Scale (RRB).
- Management for stereotypies is mostly behavioral

Pathophysiology

Foundation and developmental course of stereotypic behavior in autism is not well known[5]. Frontal white matter and both of the left and right caudate nuclei volume reduction and cortico-striatal-thalamo-cortical circuitry dysfunction is reported in children with stereotypy without autism[35]. Dopaminergic system is involved in stereotypies[36]. Basal ganglia dysfunction is correlated stereotypies in ASD. The higher right caudate and total putamen volume is associated with higher repetitive behaviors[37]. There is a relative hyperplasia of white matter in the cerebellum and brainstem in children with Down syndrome and ASD in comparison to Down syndrome only. Severity of stereotypies is associated with cerebellar white matter volume[38]. Frontal lobe volume has a positive association with stereotypies in autism[39].

Hand stereotypies without bruxism, and the other stereotypies is highly a sign of an MECP2 mutation in Rett syndrome[40]. There is a 25% positive family histories of motor stereotypies[31]. Underlying genetic abnormality for non-autistic motor stereotypies is suggested[31].

Repetitive behaviors do not differentiate high functioning autism and Asperger's disorder[41]. The association of social-communication impairments and stereotypies in literature has been exaggerated in autism[42].

Association of stereotypic behavior and response to growth hormone in adults with autism is suggested[43] and the infusion of oxytocin deceases repetitive behaviors[44]. The repetitive symptoms of ASD are associated with some executive processes including cognitive flexibility, working memory, and response inhibition, while it is not associated with executive processes of planning and fluency[45].

The rate of RRB is negatively associated with non verbal IQ while circumscribed interests are positively associated with non verbal IQ[46].

Lower hours of sleep per night predict stereotypic behavior in autism[47]. Sensory and social reinforcers maintain stereotypy[48].

Co-morbidities

The co-morbidity of stereotypies with tics, obsessive-compulsive behaviors, and attention

Table 3: The most frequent co-morbidities of stereotypies

Tic disorder
Obsessive compulsive disorder
Attention deficit hyperactivity disorder
Learning disorders
Catatonia

deficit hyperactivity disorder (ADHD) (25%) and learning disabilities (20%) is very high (Table 3)[7].

In a study, nearly 50% of typically developing children with motor stereotypies had ADHD (30%), tics (18%), and obsessive-compulsive behaviors/obsessive-compulsive disorder (10%) [31]. Tic is a stereotyped repetitive involuntary movement or sound[49]. The presence of repetitive behaviors is correlated with hyperactivity in autism[50]. Restrictive or repetitive behaviors in autism are related with obsessive compulsive symptoms in parents of afflicted individuals[51].

Family history of stereotypies in children is 25%, tics 33%, ADHD 10%, and mood-anxiety disorder 8%[10].

Cognitive level is a moderator for expression of stereotypic behaviors in individuals with intellectual disability but not for the autism[52].

Repetitive behaviors predict the presence or absence of autism at a high rate of diagnostic accuracy in adults with intellectual disability[53].

A loss of skill and regression in autism is associated with slightly higher repetitive behaviors[54]. Finally, 17% of older adolescents and adults with autism may have severe catatonic-like symptoms[55]. Of course, association and relationship of stereotypies and catatonia should be studied in future studies[55].

Whom to Investigate

Tics, obsessive compulsive behaviors, unusual sensory responses, social communication difficulties, rhythmic behaviors of sleep, and epileptic automatisms should be differentiated from motor stereotypies (Table 4)[7].

Of course, sometimes tic, compulsive behaviors, and automatism occur with stereotypies. EEG may help to differentiate seizure from stereotypies.

Autism and obsessive compulisive disorder can be differentiated by types of current repetitive thoughts and behavior.

Thoughts with contamination, sexual, religious, and symmetry, content and behaviors of cleaning, checking, and counting are less likely observed in autism than in obsessive compulsive disorder[22]. Children with OCD focus more than children with ASD on routines and rituals.

Obsession and compulsions in children with OCD are more sophisticated than those with ASD[56]. Some of the points that may help to differentiate stereotypies from other problems are mentioned in the Table 2.

Repetitive behavior also occurs in mental retardation[57]. However, autistic individuals more than those with mental retardation show greater severity and higher number of topographies of stereotypy and compulsions[8].

Table 4: Some key points that may help to differentiate stereotypies from other problems

Lack of presence in sleep[10]
Lack of individual's concern for the movement
Early age of onset (under 3 years) (tics onset is usually in 6–7 years)
The movements are more constant and fixed (than tics)
Involvement of different parts of body such as arms, hands, or the entire body (tics sometimes are less complicated such as eye blinks, and shoulder shrugs
More rhythmic than tics
Stereotypies are more suppressible than tics
Duration of stereotyped movements are longer and more rhythmic than tics(58)
Lack of loss of consciousness[7]
Stopping of movement abruptly by distracting or cueing of the individual(7)
Possible immediate return of the movement after stopping
Lack of premonitory urge to do the movements
Sometime it is pleasurable
Lack of 'inner tension' for suppressing of movement
Stereotypic movements can occur when the child is in activity such as playing computer game
Distraction decreases stereotypies more than tics[59]

Asperger syndrome and high functioning autism cannot be differentiated regarding repetitive behavior[60].

Outcomes

The three domains of autism improve and this improvement is not associated with age and cognitive function level[61]. This improvement is part of a 'natural history' of the development problems[61]. However, remission will not happen for the majority of children with autism[62]. Even in the children with improved language ability, the symptoms of autism were not fading. Severity of repetitive behaviors at the first assessment was in association with severity of autism symptoms and pragmatic competence at later ages[2].

The outcome of stereotypies is not clear[7] and it is usually chronic[10]. Motor stereotypies especially arm/hand movements types are chronic[31]. The severity and frequency of repetitive behaviors in ASD decrease with increase of age[63]. A study reported that it did not change in 50%, and worsened in 13%[10].

Complexity of motor repetitive behaviors in children with autism increases with the increase of age and higher IQ[9]. However, it is not clear whether all stereotyped behaviors need to be treated because some of them do not interfere with explorative and cognitive activities[64].

How to investigate children

Stereotypies can be assessed using the recently developed 43-item questionnaire of Repetitive Behavior Scale-Revised (RBS-R) (Bodfish et al, 2000). Its validation has been confirmed in children with autism spectrum disorders[8,65].

Repetitive and Restricted Behaviour Scale (RRB) is another recently introduced scale for assessment of stereotypies in autism spectrum disorders[66]. RRB includes 35 items that cover whole range of stereotypies. The degree of expression of each behavior is evaluated according to a five-level rating. RRB has been recently translated and back translated into English by author (A. G.). We are studying its Farsi version of psychometric properties. Some items of the RRB are:"repetitive body rocking", "bizarre gait", and "play and leisure rituals". Direct observation, video analysis, and motion sensors may be considered for RBB evaluation (Table 5).

Management and treatment

Management for stereotypies is mostly behavioral[55]. The role of medications for treatment of motor stereotypy disorders in typically developing children is not clear and behavioral therapy can be beneficial (Figure 2)[7,67].

Positive outcomes were usually reported after behavioral interventions such as "mechanical restraints alone or with other intervention variables", "response blocking alone or with other intervention variables", "non-contingent stimulation", "various contingency manipulations", and "microswitch clusters"[67].

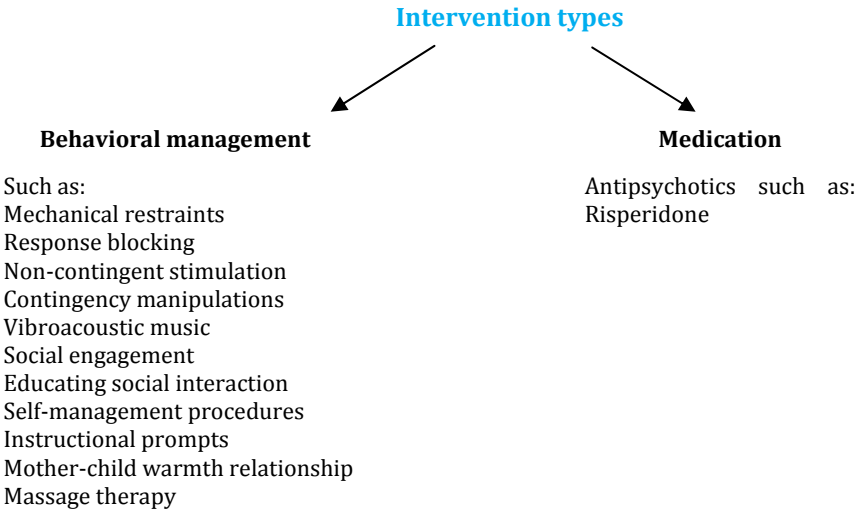
Habit reversal and differential reinforcement of other behavior improve stereotypic behaviors in non-autistic children[68].

Vibroacoustic music decreases stereotypic behaviors in individuals with autism and developmental disabilities[69]. The peer-mediated intervention and social engagement and educating social interaction decrease stereotypic behavior of children with autism[70,71]. Self-management procedures consisting of self-assessment, self-recording, and self-reinforcement decrease stereotypic behaviors in autism[24]. There is a report that

Table 5: Different methods for assessment of stereotypes

Validated standardized parent reported questionnaires
Direct observation
Videotapes analysis
Motion sensors

Fig 2: Management for stereotypies



improvement in sleep improves repetitive behaviors in autism spectrum disorder[72].

Instructional prompts reduce time spent in stereotypies[73]. Mother-child warmth relationship reduces repetitive behaviors in autism[74]. Antecedent aerobic exercise decreases stereotypic behaviors [75]. There is a report that massage therapy improves autism[76].

A randomized clinical trial showed that medication plus parent training reduces stereotypies in ASD more than medication alone[77]. The results of studies for medication management for RRB in autism are mixed[78].

Antipsychotics reduce stereotypies[79].

Risperidone may improves some sensory problems such as hyperacusia in children with autism[80]. Double-blind, placebo-controlled trial studies indicated that risperidone improves the restricted, repetitive, and stereotypic behavior of autistic children[81,82]. The synergistic effect of combination of risperidone and pentoxifylline improves behavioral problems and stereotypies in autism[83].

Stimulants reduce hyperactivity and improve attention but they may increase stereotypies[79].

Another randomized, placebo-controlled, crossover study of methylphenidate for ADHD symptoms in preschoolers PDD or intellectual disability (ID) indicated that methylphenidte increased stereotypic behavior in half of children[84]. However, another double-blind crossover study using placebo and two mehylphenidate doses did not indicate worsening stereotypic movements[85]. Secretin does not show any benefit in autism. Alternative treatments have not shown efficacy in well-designed studies[79].

Serotonin reuptake inhibitors (SRIs) such as fluvoxamine and serotonin non-specific reuptake inhibitor of clomipramine improve repetitive behavior in autism[86]. An open-label investigation in adults with autism indicated that sertraline is effective for treatment of their repetitive and aggressive symptoms[87]. A double-blind, placebo-controlled study of fluvoxamine in adults with autism reported that repetitive thoughts and behavior were decreased[88]. Meanwhile, a randomized controlled trial indicated that citalopram was not effective to decrease repetitive behavior in children with ASD[89]. Divalproex is suggested for treatment of repetitive behaviors in ASD[90].

The efficacy of naltrexone on stereotypic behavior in children with ASD was not confirmed in double-blind placebo-controlled trials[91,92]. Implications for electroconvulsive therapy in children with ASD for management of some behavioral problems is encouraged[93].

Conclusion

The key symptom of stereotypic behaviors is related to severity and progression of ASD.

Intervention programs should be undertaken in early ages because stereotypic behaviors impact on later ages and its early detection plays an important role in its management. Considering the classification system directs clinicians for future decision making. Sometimes its management is part of an underlying disease or common co-morbid conditions such as tics, autism and mental retardation. So, sometimes wider assessment may be required to prevent misdiagnosis or incorrect treatment[94]. Its management is most likely behavioral interventions.

The effect of pharmacotherapy on stereotypic behaviors is not well studied. However, anti-psychotics such as risperidone may improve it.

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CME QUESTIONS

1. Which one is not a characteristic of motor stereotypies in children with autism?

a) Suppressible
b) Repetitive
c) Rhythmicity
d) Purposeless

2. Stereotypies could be seen in which of the following conditions?

a) Developmental delay
b) Sensory deprivation
c) Autism
d) All of the above mentioned conditions

3. Which one is incorrect about epidemiology of stereotypic movement:

a) It usually starts before age 3 years
b) Boys more than girls are afflicted
c) Its rate in children with autism is less common than typically developing children
d) More than two thirds of children with autism have at least one subtype of stereotypy

4. Which one is preferred for the management of stereotypies?

a) Behavioral
b) Psychodynamic approach
c) Medication
d) Cognitive therapy

5. Which one should be differentiated from stereotypies?

a) Tics
b) Obsessive compulsive behaviors
c) Rhythmic behaviors of sleep
d) Epileptic automatisms
e) All