

Review Article

Developmental regression in autism spectrum disorder

Nouf Backer Al Backer

Department of Paediatrics, College of Medicine,
King Saud University and King Khalid University Hospital,
Riyadh, Saudi Arabia

ABSTRACT

The occurrence of developmental regression in autism spectrum disorder (ASD) is one of the most puzzling phenomena of this disorder. A little is known about the nature and mechanism of developmental regression in ASD. About one-third of young children with ASD lose some skills during the preschool period, usually speech, but sometimes also nonverbal communication, social or play skills are also affected. There is a lot of evidence suggesting that most children who demonstrate regression also had previous, subtle, developmental differences. It is difficult to predict the prognosis of autistic children with developmental regression. It seems that the earlier development of social, language, and attachment behaviors followed

by regression does not predict the later recovery of skills or better developmental outcomes. The underlying mechanisms that lead to regression in autism are unknown. The role of subclinical epilepsy in the developmental regression of children with autism remains unclear.

Key words:

Autism spectrum disorder; Developmental regression; Language regression; Social attachment.

Correspondence to:

Dr. Nouf Backer Al Backer

Assistant Professor of Paediatrics,
Consultant, Developmental Behavioral Paediatrics
Department of Paediatrics (39),
College of Medicine and King Khalid University
Hospital,
King Saud University, P.O Box 2925,
Riyadh 11461, Saudi Arabia.
Telephone: +966 554180080
Fax: +966(11) 467-9463
E-mail: nalbacker1@yahoo.com

How to cite this article:

Al Backer NB. Developmental regression in autism spectrum disorder. Sudan J Paediatr 2015; 15(1): 21 - 26.

INTRODUCTION

Autism spectrum disorders (ASD) represent a heterogeneous group of developmental disabilities with core features of social and communication impairments alongside circumscribed interests and/or repetitive motor behaviours. Individuals with ASD commonly present with a variety of associated conditions including epilepsy [1], behaviour issues [2], anxiety [3], impaired cognitive abilities [4], and loss of skills in early childhood [5].

Developmental regression in autism is one of the most challenging features of this disorder. Although several studies have documented the validity of parental reports of regression using home videos, however, data suggests that most children who demonstrate regression also demonstrated previous, subtle, developmental differences.

Autism is the most frequent condition in which regression occurs. Another disorder that demonstrates an early regression with no known aetiology is the childhood disintegrative disorder, which demonstrates behavioural relations to autism. In addition to this, another two biological conditions with known etiologies also involve regression with some behaviors resembling autism behavioral phenotype. These are Rett syndrome (a genetic disorder) and Landau-Kleffner syndrome (which involves a seizure disorder). The primary goals of this paper are to provide an overview of the various operational definitions of the term “regression” in the literature. This includes the five broad operational definitions of regression namely; language, social, language + social, mixed, and unspecified onset of regression; and characteristics of children who have experienced such a regression.

The literature regarding ASD with regression (ASD-R) has grown significantly over the past 25 years. There is no universally agreed definition; however, all definitions support that regression involves the loss of a previously attained skill, such as language.

Parent reports and clinical observation have led to a general acceptance of at least two distinct patterns of early development in autism. The most commonly reported is a gradual course of onset in which certain behaviors or their lack cause parental concern during the child’s first 2 years. This results in a diagnosis being given between 3–4 years of age [6,7]. Often, parents report failure to notice symptoms until the second year of life, however; with hindsight they realize that behavioral abnormalities were present much earlier [8]. The second pattern of early course is characterized by normal or near-normal development followed by a loss of skills during the first or second year of life [9-12].

At present, two specified classes of the term regression are evident in the autism literature, language, and language/social regressions [13]. Language regression refers, to the loss of verbalizations while language/social regression, indicates other social behavior involved in addition to the language. From these operational terms, an argument may be made for the existence of social regression independent of language regression; social regression alone is extremely rare. A number of researchers reported other non-defined regressions (e.g., “autistic regression”) or loss of other nonlanguage or nonsocial regressions (e.g., “motor regression”). These are collectively referred to here as mixed regression, but most cases appear to reflect the loss of verbalizations primarily (i.e., language regression).

It was thought that the loss of skills was the typical onset pattern in autism. However, the research data demonstrate that regression is characteristic of an only minority of children.

ETIOLOGY

To date, the causes of regression in autism are unknown. Potential factors, such as epilepsy [14], epileptiform EEGs [12] and early childhood

immunizations [15-17] have not supported causal, or even correlational linkages.

Potentially promising areas indicate that a familial history of autoimmune thyroid disease may be implicated in ASD-R development [17] and initial genes have been located. Also, the roles of different psychosocial stressors are not well-established yet [18].

CLINICAL PRESENTATION

Timing of regression

The timing of the onset of ASD-R is of interest to clinicians and parents, because of the importance of early intervention. A number of studies have reported data on the timing of regression ranging anywhere from the second year of life to 81 months [19]. A recent meta-analysis found that across 28 studies, the average reported age of regression was around 20 months of age [20]. Several researchers have reported data regarding the proportion of children with ASD-R whose onset of skill loss falls into certain pre specified age ranges instead of averages [21]. These data indicate that a minority of parents report their child lost skills after the second year of life. Overall, regression typically occurs within the second and third years of life with a mean occurrence of around 20 months. The nature of the study group should be taken into consideration when reporting percentage of children with regression.

Behavioural changes

Loss of language is the most frequently reported aspect of regression by parents [18]. Language loss occurs in children who have a very limited verbal repertoire. Kurita [10] reported that 94% of children with autism and speech loss had only single-word speech (and an extremely limited vocabulary) at the time of regression. The social skill loss was also extensive, involving loss of eye contact, as well as loss of social

interests and imitative games. A minority of ASD-R lost motor skills and basic adaptive skills, such as self-feeding and toileting [18].

Characteristics of children who have experienced regression

No relationships have been identified between autistic regression and any characteristic family feature. Differences in socioeconomic status, ethnicity, birth order, high-risk birth events, gender and age at diagnosis are not associated with regression [18,22].

Regression and core autism symptomatology

A lot of evidence has supported that children with mixed and language ASD-R have higher levels of autism symptomatology than children with ASD-NR [15,23,24]. Similarly, children with language regression [25-27] score higher on autism symptomatology when compared to children with ASD-NR. The most likely source of these differences is from greater social communication impairments in children with language regression.

Correlates and prognosis for children experiencing regression:

ASD-R and cognitive ability:

Data indicate that IQ averages are comparable to children with ASD-R and ASD-NR. However, a difference is found when considering the proportions of individuals in each group whose cognitive abilities fall within lower cognitive ability ranges [13]. Children with language [21,22] or mixed [23,28] regression constitute higher proportions of individuals with moderate to severe intellectual difficulties compared to the ASD-NR groups. Also, individuals with ASD-R are more likely to have an intellectual disability compared to individuals with ASD-NR.

ASD-R and developmental milestones and developmental skills:

No differences were reported in regard to the daily living skills of children with ASD-R and ASD-NR using Vineland Adapted Behaviour Scales [27,29,30] or fine motor skills [29].

Recent studies indicate that development of children with ASD-R and ASD-NR does not differ in any dramatic manner, aside from earlier language attainment in children with ASD-R. The evidence is mixed regarding the age at which phrase speech is used in children with ASD-R compared to ASD-NR. However, data regarding the age of first steps [22] the general developmental, daily living skills, and fine motor skill attainment [27,29-31] indicate that children with ASD-R do not differ from ASD-NR.

CONCLUSION

Interpretation of the existing data regarding regression is complicated by the usage of multiple, often poorly defined, operational definitions. It seems that children with mixed and language ASD-R are more likely to have cognitive deficiencies compared to children with ASD-NR, as well as higher levels of autistic symptomatology, particularly in the area of social communication symptoms.

Although we do not yet understand the biology of regression in autism, we have much information about the phenomenology. It is a very painful experience for parents who search for explanations and keep blaming themselves. Developmental regression may, however, be an early and reliable marker in a number of children with autism.

REFERENCES

1. Canitano R, Luchetti A, Zappella M. Epilepsy, electroencephalographic abnormalities and regression in children with autism. *J Child Neurol* 2005; 20:27–31.
2. Dumas J, Wolf L, Fisman S, Culligan A. Parenting stress, child behavior problems, and dysphoria in parents of children with autism, down syndrome, behavior disorders, and normal development. *Exceptionality* 1991; 2:97–110.
3. Gillott A, Furniss F, Walter A. Anxiety in high-functioning children with autism. *Autism* 2001; 5:277–286.
4. Hill E. Executive dysfunction in autism. *Trends Cogn Sci* 2004; 8:26-32.
5. Stefanatos GA. Regression in autistic spectrum disorders. *Neuropsychol Rev* 2008; 18:305–19.
1. Hoshino Y, Kaneko M, Yashima Y, Kumashiro H, Volkmar FR, Cohen DJ. Clinical features of autistic children with setback course in their infancy. *Jpn J Psychiat Neurol* 1987; 41:237–245.
2. Volkmar FR, Stier DM, Cohen DJ. Age of recognition of pervasive developmental disorder. *Am J Psychiat* 1985; 142:1450 –1452.
3. Lord C, Shulman C, Di Lavore P. Regression and word loss in autistic spectrum disorders. *J Child Psychol Psychiatry* 2004; 45:936–955.
4. Harper J. Age and type of onset as critical variables in early infantile autism. *J of autism and childhood schizophrenia*. 1975; 5:25-36
5. Kurita H. Infantile autism with speech loss before the age of thirty months. *J Am Acad Child Adolesc Psychiatry* 1985; 24:191–196.

6. Rogers S, DiLalla D. Age of symptom onset in young children with pervasive developmental disorders. *J Am Acad Child Adolesc Psychiatry* 1990; 29:863–872.
7. Tuchman RF, Rapin I. Regression in pervasive developmental disorders: Seizures and epileptiform electroencephalogram correlates. *Pediatrics* 1997; 99:560–566.
8. Barger B, Campbell J. Developmental Regression in Autism Spectrum Disorders: Implications for Clinical Outcomes. In: Patel VB, Preedy VR, Martin CR, editors. *Comprehensive Guide to Autism*, Springer New York; 2014, p. 1473–1493.
9. Giannotti F, Cortesi F, Cerquiglini A, Miraglia D, Vagnoni C, Sebastianai T, et al. An investigation of sleep characteristics, EEG abnormalities and epilepsy in developmentally regressed and non-regressed children with autism. *J Autism Dev Disord* 2008; 38:1888–1897.
10. Meilleur A, Fombonne E. Regression of language and non-language skills in pervasive developmental disorders. *J Intellect Disabil Res* 2009; 53:115–124.
11. Richler J, Luyster R, Risi S, Hsu W, Dawson G, Bernier R, et al. Is there a “regressive phenotype” of autism spectrum disorder associated with the measles-mumps-rubella vaccine? A CPEA study. *J Autism Dev Disord* 2006; 36:299–316.
12. Molloy C, Morrow A, Meinzen-Deir J, Dawson G, Bernier R, Dunn M et al. Familial autoimmune thyroid disease as a risk factor for regression in children with autism spectrum disorder: a CPEA study. *J Autism Dev Disord* 2006; 36:317–324.
13. Rogers SJ. Developmental regression in autism spectrum disorders. *Ment Retard Dev Disabil Res Rev* 2004; 10: 139–143.
14. Castillo H, Patterson B, Hicker F, Kinsman A, Howard JM, Mitchell T et al. Difference in age of regression in children with autism with and without down syndrome. *J Dev Behav Pediatr* 2008; 29:89–92.
15. Barger BD, Campbell JM, McDonough JD. Prevalence of regression in autism: quantitative synthesis. *International Meeting for Autism Research*, Manchester, UK; 2012.
16. Kobayashi R, Murata T. Setback phenomenon in autism and long-term prognosis. *Acta Psychiatr Scand* 1998; 98:296–303.
17. Christopher JA, Sears LL, Williams PG, Oliver J, Hersh J. Familial, medical and developmental patterns of children with autism and a history of language regression. *J Dev Phys Disabil* 2004; 16:163–170.
18. Oslejskova H, Dusek L, Makovska Z, Pejcochová J, Autrata R, Slapák I. Complicated relationship between autism with regression and epilepsy. *Neuroendocrinol Lett* 2008; 29:558–570.
19. Stella J, Mundy P, Tuchman R. Social and nonsocial factors in the childhood autism rating scale. *J Autism Dev Disord* 1999; 29:307–317
20. Baird G, Charman T, Pickles A, Chandler S, Loucas T, Meldrum D, et al. Regression, developmental trajectory and associated problems in disorders in the autism spectrum: the SNAP study. *J Autism Dev Disord* 2008; 38:1827–1836.
21. Kalb L, Law J, Landa R, Law P. Onset patterns prior to 36 months in autism spectrum disorder. *J Autism Dev Disord* 2010; 40:1389–1402.
22. Parr JR, Le Couteur A, Baird G, Rutter M, Pickles A, Fombonne E, et al. Early developmental regression in autism spectrum disorder: evidence from an international multiplex sample. *J Autism Dev Disord* 2011; 41:332–340.
23. Wiggins LD, Rice CE, Baio J. Developmental regression in children with an autism spectrum disorder identified

- by a population- based surveillance system. *Autism* 2009; 13:357–374.
24. Chawarska K, Paul R, Klin A, Hannigen S, Dichtel LE, Volkmar F, et al. Parental recognition of developmental problems in toddlers with autism spectrum disorders. *J Autism Dev Disord* 2007; 37:62–72.
 25. Hansen RL, Ozonoff S, Krakowiak P, Angkustsiri K, Jones C, Deprey LJ, et al. Regression in autism: prevalence and associated factors in the CHARGE study. *Ambul Pediatr* 2008; 8:25–31.
 26. Ozonoff S, Heung K, Byrd R, Hansen R, Hertz-Picciotto I. The onset of autism: patterns of symptom emergence in the first years of life. *Autism Res* 2008; 1:320–328.