LANGUAGE REGRESSION IN CHILDREN WITH AUTISM SPECTRUM DISORDER

[1]Ayesha Fayyaz, [2]Faryal Ikram, [3] Muzammil Nasir, [4]Madiha Manzoor, [5]Maham Ikram, [6]Kanwal Shahzadi

1. Speech and Language Pathologist at Govt. Secondary Special Education Centre Rahim Yar Khan
Email ID: ayeshatarar6@gmail.com   (Corresponding Email unlimitedone271@gmail.com)
2. Speech and Language Pathologist In Cmh Multan, CEO of Rehman Rehabilation Center
3. Speech Language Pathologist at Dear Life Rehabilitation Center for Specially-Abled Children and Youngsters Lahore.
4. Speech Language Pathologist at Dear Life Rehabilitation Center for Specially-Abled Children and Youngsters Lahore.
5. Speech and Language Pathologist in Rehman Rehablitation Center
6. Speech and Language Pathologist in Faizan Rehabilitation Center Lahore

ABSTRACT

Regression in Autism applies to the phenomenon of apparently normal early development followed by the loss of previously acquired skills and manifestation of symptoms of autism. Estimates of frequency of language regression range from 95-99%. The study was aimed to describe frequency of language regression and mean age of language regression in children with Autism Spectrum disorder. A questionnaire was developed based on early language milestones. it comprised of 23 skills. This retrospective study was conducted on a single group. The participants consisted of parents of 67 children with ASD (male 55, female 12). 65 parents out of 67 reported language regression during early years of life. The findings revealed significant regression in children with ASD. The mean regression age is between 1-2 years. Based on the findings it can be stated that language regression is a diagnostic feature of children with ASD, and the mean age of language regression is between 1-2 years of age and help to identify disorder as early as between 1-2 years of age for early intervention.

Keywords: *Language, Regression, Autism Spectrum Disorder, Child health*

INTRODUCTION

1.1   Definition of Autism:

Autism spectrum disorder (ASD) is a neuropsychiatric disorder of primary childhood which may be defined entirely on the basis of global impairments in the child's ability to develop social skills, to communicate as well as to understand language. (Miles et al; 2005)

Autism is a developmental disorder belonging to diverse characteristics leading to marked deficits in social interaction, markedly atypical communication as well as the presence of restricted repetitive behaviors and interest. The sign in the autistic children are present from early childhood might face challenges in the activities of daily living. (Lord et al; 2015)

Autism is an umbrella term used to describe three developmental disorders such as autism, pervasive developmental disorder not otherwise specified (PDD-NOS) and asperger disorder having common symptoms and etiology. (Rapin, Tuchman 2008)

Autism is a neurodevelopmental disorder characterized by impairments in reciprocal social interaction, impairments in verbal and non-verbal communication, lack of imaginative play, and a pattern of repetitive , stereotypical behaviors and interests. (Juul-Dam, Townsend and Courchense, 2004)

Autism spectrum disorder is a heterogeneous neurodevelopment disorder which is defined clinically by characteristics behavioral impairments in :

- Reciprocal social interaction
- Verbal and non-verbal communication
- Repetitive and restricted patterns of behaviour, interests and activities (Webmed,2015)

1.1 Symptoms of Autism:

There are three main symptoms of autism:

a) in social and non-social context might be resulting in insufficient or inappropriate social interactions.

b) Inflexibility and perseveration including both stereotypic movements such as purposeless repetitive movements as well as resistance to change.

c) Impairment Marked impairment in the receptive as well as expressive language. (Maston et al., 2011)

1.2   Causes of Autism Spectrum Disorder:

Autism is really a sophisticated ailment. The causes of ASD usually are not acknowledged .Subsequent aspects may possibly play a role in ASD.

- Issues that happen in the prenatal, neonatal time periods or during delivery may become reason of ASD (obstetric complications)

- Infection (Rubella)

- Hereditary variables or family record.

- Being exposed to toxins like steer and mercury.

- Age of mothers and fathers.(Landrigan,2010)

Autism seems to be causally heterogenous disorder. In the literature there is varying support for a wide spectrum of hypotheses regarding the cause of autism. It may be due to genetic factors or enviornmental factors. But there are risk factor that increases the chance of having a child With autism. Advanced age of the mother or the father increases the chance of an autistic child. (Reichenberg *et al.*, 2006)

1.4   Diagnostic criteria for Autism:

A. Persistent deficits in social interaction and communication across multiple contexts, as manifested by the following, currently or by history.

- Deficits in social-emotional reciprocity ,ranging, for example, from abnormal social approach and failure of normal back- and –forth conversation; to reduce sharing of interests, emotions or affect ;to failure to initiate or respond to social interactions.

- Deficits in nonverbal communicative behaviors used for social interaction, ranging for example ,from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language and deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication.

- Deficits in developing, understanding and maintaining relationships, ranging for example, from difficulty adjusting behavior suit various social contexts; to difficulties in sharing, imaginative play or in making friends; to absence of interests in peers.

B. Restricted, repetitive patterns of behavior, interests and activities, as manifested by at least two of the following, currently or by history.

- Stereotype type or repetitive motor movements, use of objects, or

speech(e.g simple motor stereotypies, lining up toys or flipping objects, echolalia, idiosyncratic phrases)

- Insistence or sameness,inflexible adherence to routines ,or ritualized patterns of verbal or nonverbal behavior (e.g extreme distress at small changes, difficulties with transitions,rigid thinking patterns, greeting rituals, need to take same route or eat same food everyday)

- Highly restricted, fixated interests that are abnormal in intensity or focus (e.g strong attachement to or preoccupation with unusual objects, excessively circumscribed or perseverative interests.)

- Hyper-or hypoactivity to sensory input or unusual interest in sensory aspects of the environment (e.g apparent indifference to pain/temperature, adverse response to specific sounds or textures, excessive smelling or touching of objects, visual fascination with lights or movement.)

C.   Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities, or may be masked by learned strategies in later life)

D.   Symptoms cause clinically significant impairment in social, occupational, or the important areas of current functioning.

E.   These disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnosis of autism spectrum disorder and intellectual disability; social communication should be below that expected for general developmental level. (Deficits *et al.*, 2013)

Table no 1.1   Severity Level:

Level:1 Requiring very substantial support	Social communication: Severe deficits in verbal and nonverbal social communication skills cause severe impairments in functioning; very limited initiation of social interactions and minimal response to social overtures from others.	Restricted interests& repetitive behaviors: Pre-occupations,fixated rituals or repetitive behaviors markedly interfere with functioning in all spheres. Marked distress when rituals and routines are interrupted; very difficult to redirect from fixated interests or returns to it quickly.
Level:2 Requiring substantial support	Marked deficits in verbal and nonverbal communication skills; social impairments apparent even with supports in place; limited initiation of social interactions and reduced or abnormal response to social overtures from others.	RRBs and/or preoccupations or fixated interests appear frequently enough to be obvious to the causal observer and interfere with functioning in a variety of texts. Distress or frustration is apparent when RRBs are interrupted; difficult to redirect from fixated interest.
Level:3 Requiring support	Without supports in place, deficits in social communication cause noticeable impairments. Has difficulty initiating social interactions and demonstrates clear examples of atypical or unsuccessful response to social overtures of others.	RRBs cause significant interference with functioning in one or more contexts. Resists attempts by others to interrupt RRBs or to be redirected from fixated interests.

1.5   Behaviors/Symptoms that are not/May not be captured in DSM-5 ASD: (Deficits et al., 2013)

- Problems with play/ imagination

- Shyness/social anxiety

- Language and development delays

- Behavioral difficulties/temper tantrums

- Poor initiation skills

1.6   Associated Features Supporting diagnosis; (Deficits et al., 2013)

- Many individuals with autism spectrum disorder also have intellectual impairment or language impairment (e.g slow to talk, language comprehension behind production)

- The gap between intellectual and adaptive functioning skills is often large.

- Motor deficits are often present, including odd gate, clumsiness and other abnormal signs (e.g walking on tiptoes) Self injury (e.g head banging, biting the wrist) may occur.

- Disruptive and challenging behaviours are more common in children and adolescents with autism spectrum disorder than other disorders.

- Adolescents and adults with autism spectrum disorder are prone to anxiety and depression.

- Some individuals develop catatonic-like motor behavior.

1.7   Prevalence

In recent years, reported frequencies for Autism spectrum disorder across Pakistan have approached 1 in 59 children, with similar estimates in child and adult sample.

1.8   Language:

Language is a primarily human and non-instinctive method of communicating ideas, emotions, and desires by means of a system of voluntarily produced symbols. (Sapir, Language, 1921)

There are two main areas of language;

a) Receptive language (understanding language)

Comprehension of language.

b) Expressive language (using language)

The use of language through speech, signs or alternative forms of communication to communicate wants, needs, thoughts or ideas. (Listening, Sentences and Questions, 1973)

1.9   Language Regression:

Language regression means loss of language after language starts developing during early years of life.

Language regression in autism children usually appears relatively early before the children reach the age of two years. According to the reports from parents with autism children, about 25% start developing words between the ages of 12 and 18 months before losing the words. This is one of the helpful sign in autism diagnosis. According to a longitudinal study of toddlers conducted in 2004, this type of language regression that manifests itself after the onset of normal language is unique to autism. It is not seen in children with other developmental disabilities. (Goldberg WA 2003) Loss of previously acquired language at any age ,even if that language only includes a few words or communicative gestures ,is often associated with a more global regression in cognition or behavior and has serious implications for future function. (Elsevier science,2001)

1.10   Intervention to support individuals with Autism:

Researchers have found that children having autism frequently experience difficulties in different areas like socialization, communication as well as cognitive functioning leading to deficits in attending imitating, regulating and understanding the auditory stimuli.(Hayes et al.,2010)

1.11   Rationale of Study

 How to calculate frequency of language regression in children with Autism in Pakistani population and mean age of language regression?

1.2   Objective

To access frequency of language regression in children with ASD and mean age of language regression

2.   LITERATURE REVIEW

The study was aimed to describe the differences in language regression between children with ASD and typically developing children and also to determine the age of regression Form (Goldberg et al., 2003). The skills were validated by five

Clinical Psychologists. It comprised of 16 skills which included domains like, 'spoken language and non verbal communication', 'social interest and responsiveness' and 'play and imagination'. This retrospective study was conducted on a single group. The participants consisted of parents of 30 children with ASD (22 males and 8 females). Results: The findings revealed a significant regression in children with ASD. The mean regression age is 20.19 months (SD-5.2). The regression profile of the children with ASD revealed regression of language skills occurred at 19.16 months followed by non language skills at 20.5 months.(Kumar, Karmakar and Mohanan, 2014)

This study was conducted to examine the prevalence and predictors of language attainment in children with autism spectrum disorder (ASD) and severe language delay. Data used for the current study were from 535 children with ASD who were at least 8 years of age (mean = 11.6 years, SD = 2.73 years) and who did not acquire phrase speech before age 4. Logistic and Cox proportionate hazards regression analyses examined predictors of phrase and fluent speech attainment and age at acquisition, respectively. A total of 372 children (70%) attained phrase speech and 253 children (47%) attained fluent speech at or after age 4. It is concluded that many severely language-delayed children in the present sample attained phrase or fluent speech at or after age 4 years.(Wodka, Mathy and Kalb, 2013)

This study investigates correlation of language regression for children diagnosed with autism spectrum disorders (ASD). Using archival data, children diagnosed with ASD (N = 114, M age = 41.4 months) were divided into four groups based on language development (i.e., regression, plateau, general delay, no delay) and compared on developmental, adaptive behavior, symptom severity, and behavioral adjustment variables. Few overall differences emerged between groups, including similar non-language developmental history, equal risk for seizure disorder, and comparable behavioral adjustment. Groups did not differ with respect to autism symptomatology as measured by the Autism Diagnostic Observation Schedule and Autism Diagnostic Interview-Revised.(Jones and Campbell, 2010)

This study examined the occurrence of regression in 135 children with PDD, mean age 6.3 years. The sample was composed of 80 (59.4%) children diagnosed with autism, 44 (32.6%) with pervasive developmental disorder-not otherwise specified (PDD-NOS) and 11 (8%) with Asperger syndrome. The Autism Diagnostic Interview Revised (ADI-R) was used to evaluate the type of loss and to characterize associated factors including birth rank, gender and thimerosal exposure through vaccination. A

total of 30 (22%) subjects regressed: nine (30%) underwent language regression alone, 17 (57%) lost a skill other than language and four (13%) lost both language and another skill. Significantly higher levels of regression were found in autism (30%) compared with PDD-NOS (14%) and Asperger syndrome.(Meilleur and Fombonne, 2009)

This study report rates of regression and associated findings in a population derived group of 255 children aged 9-14 years, participating in a prevalence study of autism spectrum disorders (ASD); 53 with narrowly defined autism, 105 with broader ASD and 97 with non-ASD neurodevelopment problems, drawn from those with special educational needs within a population of 56,946 children. Language regression was reported in 30% with narrowly defined autism, 8% with broader ASD and less than 3% with developmental problems without ASD. A smaller group of children were identified who underwent a less clear setback. Regression was associated with higher rates of autistic symptoms and a deviation in developmental trajectory. Regression was not associated with epilepsy or gastrointestinal problems.(Baird *et al.*, 2008)

The aim of this study was to examine the prevalence of regressive autism and associated demographic, medical, and developmental factors by using 2 different definitions of regression based on the Autism Diagnostic Interview, Revised. Subjects were aged 2 to 5 years, with autism (AU) or autism spectrum disorder (ASD) confirmed by standardized measures. Children with regression, defined as a(loss of both language and social skills or b) loss of either language or social skills, were compared with each other and to children with AU or ASD with no reported loss of skills on developmental and adaptive functioning. Parents reported on seizure, gastrointestinal, and sleep concerns. Fifteen percent (50/333) of the combined AU-ASD group lost both language and social skills; 41% (138/333) lost either language or social skills. No differences were found between the 2 samples of children with regression.(Hansen *et al.*, 2008)

This systematic study conducted by Rhiannon J.Luyster, Mary Beth Kadlee …in 2008, in the state of Massachusetts. The goal of this study was to systematically investigate toddlers with ASD and to identify early correlates of expressive and receptive language in this population. Study includes 164 toddlers with ASD between the ages 18 and 33 months. Results suggested good agreement among different

measures of early language. These findings have important implications for intervention programs targeting this population.   (Luyster *et al.*, 2008)

This study report demographic and clinical characteristics of children reported to the US Vaccine Adverse Event Reporting System (VAERS) as having autism or another developmental disorder after vaccination. We completed 124 interviews with parents and reviewed medical records for 31 children whose records contained sufficient information to evaluate the child's developmental history. Medical record review indicated that 27 of 31 (87%) children had autism/ASD and 19 (61.3%) had evidence of developmental regression (loss of social, language, or motor skills). The proportion of VAERS cases of autism with regression was greater than that reported in population-based studies, based on the subset of VAERS cases with medical record.(Woo *et al.*, 2007)

This study conducted by Luyster is a multisite study of 351 children with autism spectrum disorders, 21 children with developmental delays, and 31 children with typical development, this study used caregiver interviews (i.e., the Autism Diagnostic Interview-Revised) at the time of entry into other research projects and follow-up telephone interviews designed for this project to describe the children's early acquisition and loss of social-communication milestones. Children who had used words spontaneously and meaningfully and then stopped talking were described by their caregivers as showing more gestures, greater participation in social games, and better receptive language before the loss and fewer of these skills after the loss than other children with autism spectrum disorders. A significant minority of children with autism without word loss showed a very similar pattern of loss of social-communication skills, a pattern not observed in the children with developmental delays or typical development.(Luyster *et al.*, 2005)

This study conducted by Goldberg introduced an instrument, the Regression Supplement Form, intended to supplement the Autism Diagnosis Interview-Revised and yield precise information about the types and timing of regression and events concurrent with loss and regain of skills. Data were collected from parents of 44 children (38 male, 6 female; mean age = 6 years) with Autistic Spectrum Disorder (37 Autistic Disorder, 7 Pervasive Developmental Disorder-Not Otherwise Specified). Parental responses on the Autism Diagnosis Interview-Revised indicated loss of skills during early development. The profile of regression that emerged included loss of skills between 18 and 21 months, on average, with language-only regression less

common than loss of other, nonlanguage skills only or of full regression (loss of language and other skills).(Goldberg *et al.*, 2003)

Charman conducted this study in which Parents report data on early language development measured using the MacArthur Communicative Development Inventory (CDI–Infant Form) was collected on 134 preschool children with autism spectrum disorder. The pattern of development of understanding of phrases, word comprehension and expression, and production of gestures, was compared to the typical pattern. In common with typical development there was considerable variability in language acquisition, although for the group as a whole this was significantly delayed compared to the normal course. In addition, atypical patterns were identified in the emergence of language skills in the sample. Comprehension of words was delayed in comparison to word production, and production of early gestures (involving sharing reference) was delayed relative to production of later gestures (involving use of objects).(Charman *et al.*, 2003)

The aim of this study was to observe language regression both in autistic regression and as part of acquired epileptic aphasia (Landau-Kleffner Syndrome). We prospectively identified 177 children with language regression at four major medical centers, and their clinical characteristics were recorded. Their mean age at regression was 22.8 months. The mean time-to-specialist referral was 38 months of age. Most children (88%) met criteria for autism or manifested autistic features. Males (P = 0.02) and children less than 3 years of age who regressed (P = 0.016) had a higher probability of developing autistic behaviors. Seizures were more common in children who regressed after they reached 3 years of age (P < 0.001), and children with seizures were less likely to have associated autistic regression (P < 0.001).. At last follow-up, language function was impaired in 88% of the children, although some improvement was noted in 57%. We conclude that the loss of previously acquired language at any age, even if that language only includes a few words or communicative gestures, is often associated with a more global regression in cognition and/or behavior and has serious implications for future function.(Shinnar *et al.*, 2001)

3.   MATERIALS AND METHODS

3.1 Study Population

 Parents of Children with ASD age ranging from 3-6 years.

3.2 Study Design:

This is an observational study.

3.3 Study Settings:

Department of Developmental Pediatrics Children's Hospital Lahore, School of Allied Health Sciences.

3.4 Duration of Study:

Six months after approval of synopsis.(July 2019 to Dec 2019)

3.5 Sample Size:

The sample size was calculated using formula below;

$$n = \frac{z^2 p(1-p)}{d^2}$$

$$n = \frac{(1.96)^2 0.016(1-0.016)}{(0.03)^2}$$

$$n = 67$$

N=sample size

Z= level of confidence (1.96 for 95% confidence level)

P= prevalence or proportion taken as(0.016 according to latest research)

3.6 Data Collection Tool:

Questionnaire is used.

3.7 Operational Definition of Variables:

Autism:

Autism spectrum disorder is pervasive developmental disorders, characterized by three core deficits i.e...Social interaction and communication problems and restricted, stereotype, and repetitive behaviors.(Freitag, Kleser and Von Gontardf, 2006)

Language Regression:

Language regression means loss of language after language starts developing during early years of life.(Goldberg WA 2003)

3.8 Data Collection Procedure:

All the patients fulfilling their inclusive criteria visiting ASD team in the Department of Developmental pediatrics were evaluated by the speech therapist, and investigated for the frequency of language regression and its mean age. We consider newly diagnosed cases of autism. When we ask parents of autistic children about the language development of their children, they were quite upset and depressed about the problem of their children and were curious to know why this happen to their children. Most of the informants were mothers of autistic children. We gave complete or detailed description about the purpose of the study and how it will be beneficial for society. Questionnaire based on 23 questions filled by the them and purpose of the study was explained time to time when needed.

3.9 Sampling Criteria:

Inclusion criteria

- Children with autism spectrum disorder, age ranging from 3-6 years.
- Newly diagnosed cases of autism.
- Both male and female.

Exclusion Criteria

All other patients who are not fulfilling the above-mentioned inclusion criteria are excluded.

3.10 Ethical Issue:

There is no ethical issue because it's an observational study and no therapy is applied.

3.11 Statistical analyses:

The data was entered using **IBM-SPSS v-23,** the continuous variables like age are Expressed in the form of Mean ± SD and categorical variable in frequency and proportions. Graphs were used to display the data.

4. RESULTS

This observational study based on six months duration and carried out at Department of Developmental & Behavioral Pediatrics Children Hospital & Institute of Child Health Lahore. This research was done to know the frequency of language regression and mean age of language regression in autistic individuals.67 autistic children participated in this research, Age ranging from 3-6 years,55 were male and 12 were females.

4.1 Descriptive statistics of age of study sample

	N	Minimum	Maximum	Mean	Standard deviation
Age	67	3.0	5.10	4.083	0.7127

Table 4.1 shows descriptive statistics of age of study sample. Minimum age of autistic children is 3 year and maximum age is 5.10 years with mean of 4.083 and standard deviation .7127 .

Table 4.2 frequency distribution of Gender

Male	55	82.1%
Female	12	17.9%

Table 4.2 shows frequency of male to female ratio. out of 67 participants 55 were males and twelve were females and the ratio between autistic males and females is almost 4:1
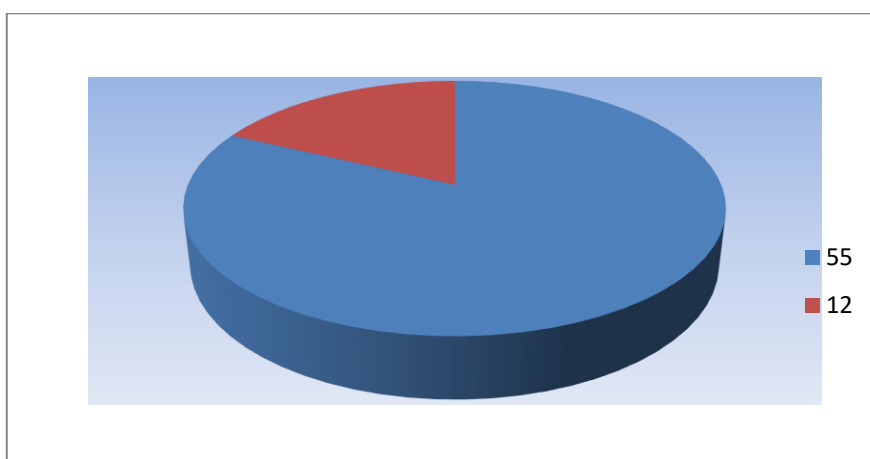

Figure 1

Table 4.3 Frequency distribution of Birth order

1	47	70.1%
2	16	23.9%
3	2	3%
5	2	3%

Table 4.3 shows frequency of birth order in autistic children, out of 67 children 47 children have 1st birth order,16 have 2nd birth order,2 have 3rd and 2 have 5th birth order. Almost 70% children were the elder or first babies of their families. And this disorder is least common in 2nd or 3rd birth order children.
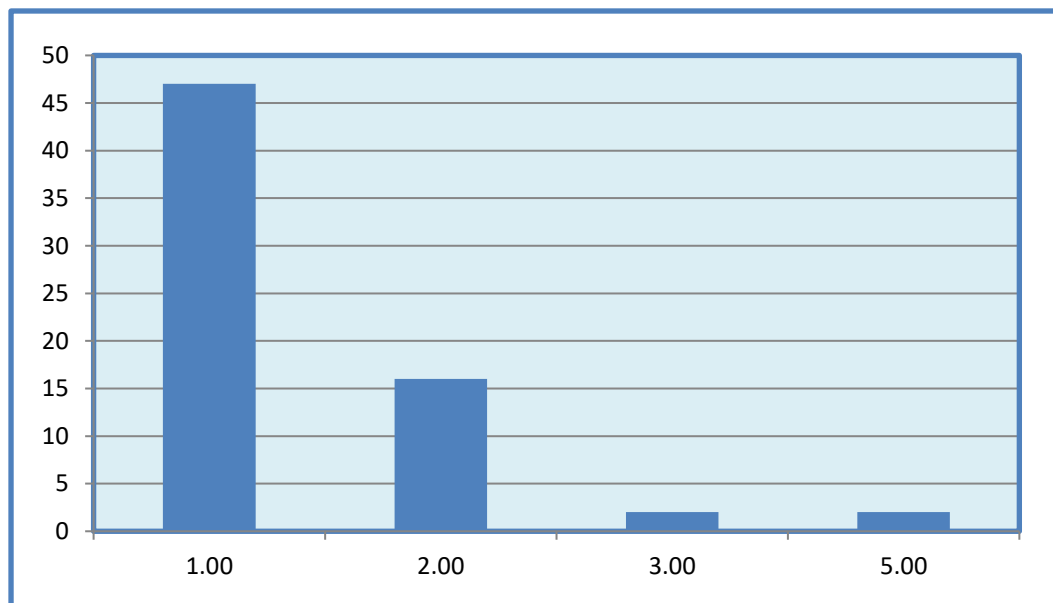

Figure 2

Table 4.4 Frequency of Language variables

	No	Yes
Coos and gurgles when content	1	66
Babbles series of syllables	1	66
Smiles in response to expressions of others and mirror image	4	63
peekk-a-boo,clapshands,and waves bye-bye in immitation of adults	6	61
Says mama-dada with meaning	20	47
Knows afew body parts and point	42	25
Follow a simple one step directions	54	13
Vocabulary more than 50 words	62	5
Uses some one or two word questions	64	3
Puts two words together	63	4
Vocabulary of 400 words	66	1
Uses 2-3 word phrases	67	0
Knows age and sex and counts three objects correctly.	67	0

Table 4.4 indicates frequency of different variables including cooing (mean=.9851), babbling(mean=.9851) smiles (mean=.9403), peek-a-boo(mean=.30819), says mama-dada with meaning(mean=.7015), knows a few parts of the body and point(mean=.3731) follow simple one step direction(mean=.1940),vocabulary more than 50 words(mean=.0746) , use some one or two word questions(mean=.0448), puts two words together(mean=.0597),vocabulary of 400 words(mean.0000),uses 2-3 word phrases(mean=.0000),.and this table shows regression from cooing to 2-3 word phrases. Majority of Autistic children regress between 1-2 years. Parents reported that

children were developing normally during early phase of life and suddenly shows regression both in language and social skills.
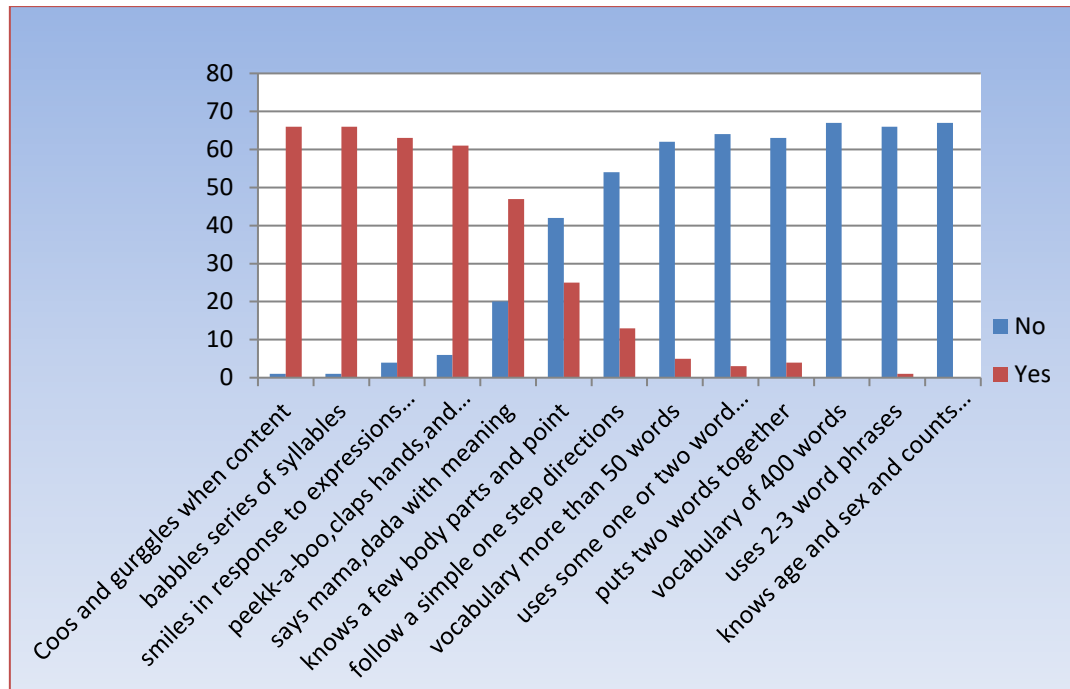

Figure 3

5. DISCUSSION

Though several studies have been done on language regression of children with ASD however, relatively few studies detailed the nature of this loss. When the age of regression was reported most of the children have progressed beyond the stage of complex babbling and proto-word production, and are functioning at single word level, with emerging or expanding expressive vocabularies of words that they use spontaneously and meaningfully. During the course of regression, children cease to produce words that previously seemed a permanent part of their vocabulary while others showed marked reduction in their verbalizations, often to the point where they discontinue speaking altogether.

Kurita [30] and Rutter [33] stated that around 20-40% of children, who had regression, lost all expressive language. Although regression age of language domain

was between 1-2 years in present study however large scale study done by Kurita found 37.2% of 261 children lost speech preceding to 30 months of age.

Wilson et al.[21] stated that the parents of children with autism reported language regression between 18 and 24 months of age and 95% before age 3 years which was also similar to current findings.

Goldberg stated that the profile of regression that emerged included loss of skills between 18 and 21 months, on average, with language only regression less common than loss of other, nonlanguage skills only or full regression(loss of language and other skills).

Woo reported that 27 of 31(87%) children had autism and 19(61.3%) had evidence of developmental regression (loss of social, language and motor skills).

The findings of the present study revealed that 65 of 67 children (97%) with Autism spectrum disorder with age ranging from 3-6 years have language regression and the mean age of language regression was between 1-2 years of age. Several reports have suggested that most children who regress in language demonstrated at least mild to moderate regression in social skills, evident in poor eye gaze, loss of social smiling, loss of interests in others, withdrawn or aloof behavior and problems involving play skills.

5.1   Limitations:

- One of the limitation of the study was shortage of time
- The study population was taken from only one institute.
- Sample size was not enough.

5.2   Suggestions:

- Increase time of study.
- Increase sample size.
- Other institutes should include.

5.3   Implications:

It will be helpful for the researcher, speech and language pathologists and psychologists if they want to use for their purpose.

5.4   Conclusions:

Based on the findings it can be stated that language regression is a diagnostic feature of children with ASD, and the mean age of language regression is between 1-2 years of age and help to identify disorder as early as between 1-2 years of age for early intervention.

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