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Developmental Regression in Autism: Maternal Perception

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Developmental regression among children with autism is a common phenomenon of unknown origin. The purpose of this study was to identify the differences between children with autism who reportedly regressed with those who did not regress. A representative group of 39 mothers were interviewed (40 children—1 pair of twin girls) about familial, pregnancy, perinatal, as well as medical history and developmental milestones. The study focused on mothers' perceptions of developmental regression. Nineteen children (47.5%) regressed in verbal and nonverbal communication and social but not in motor abilities. Mean age of regression was 24 months, with 11 children who regressed before and 8 after this age. No significant differences were reported by mothers of children who did or did not regress. More mothers of children who regressed, than those of children who did not, expressed guilt feelings regarding the development of autism, and almost all of them had an "explanation" for the possible mechanisms that might have influenced their children's developmental course. In conclusion, developmental regression in our population appears to be a typical event in the natural course of autism. There is little difference between those children who regressed and those who did not regress in maternal perceptions and reports of development, family, and medical history.

KEY WORDS: Autism; developmental regression; maternal reports.

INTRODUCTION

Developmental regression is a common observation among children with autism. To date, different prevalence rates between 20 and 49% have been reported (Hoshino *et al.*, 1987; Kurita 1985; Rutter & Lord, 1987). Regression in language, social skills, and play is usually observed between the first and the third year of life (Tuchman & Rapin, 1997). Some parents perceive their children as having typical development, whereas others report an abnormal acquisition of developmental milestones, prior to the regression. Hoshino *et al.* (1987) reported that children with autism who regressed had more severe speech problems, more severe behavioral disorders, and lower adaptive levels at schools or institutes than children with autism who experienced no regression. Autistic children who lost their expressive verbal ability had significant lower IQs than children who did not loose speech (Rogers & Dilalla, 1990). Similarly, children who regressed were perceived by their parents to use oral communication less well than their autistic peers who did not regress (Brown & Prelock, 1995). However, in the same study, most children who did not demonstrate verbal communication showed no regression in early childhood. Kobayashi and Murata (1998) also reported lower level of language development among children with autism who regressed, as compared to children who had not regressed, upon entering elementary school. However, there was no difference between the groups in their levels of adaptive skills

The underlying mechanisms that lead to the regression in autism are unknown. Epileptic seizures, which

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could possibly account for a regressive course, are reported in only a third of the children and adults with autism (Volkmar & Nelson, 1990). There is controversy regarding the frequency of epilepsy in children who regress in comparison with children who do not regress. (Kobayashi & Murata, 1998; Tuchman & Rapin, 1997). Neither specific physical, neurological, nor other positive medical findings were reported for regressed groups when compared with children who did not regress. However, regression is significant negative change in the child's condition. This serves as a signal to the parents of their child's deterioration, and may result in increasing parental motivation to seek help at an earlier stage. Regression is usually reported in a wide age range from 15–30 months (Hoshino *et al.*, 1987). In our clinical practice we have noted that more regressed children are presented at early ages (around 18 months) and fewer children are reported as regressed at later ages.

In Israel, all children with developmental disorders are seen for evaluation and/or treatment at public medical centers. The prevalence of children who are diagnosed as autistic is similar to the prevalence reported in recent literature (Davidovitch *et al.*, in preparation). Since it is possible to identify all children diagnosed within a particular time range, this gave us the opportunity to compare and contrast a total population of autistic children who have regressed with those who have not. We were also interested in comparing children who regressed early with children who regressed at a later stage of development.

This study had several goals. One was to delineate differences between children whose mothers reported regression with those whose mothers did not report regression. In addition, we wished to compare children who were reported as regressing before 24 months (Early) with children who had been reported as regressing after 24 months (Late), in an attempt to discover differences between these two subgroups. Late regression is more dramatic and severe. Establishing subgroups for comparison may lead to better understanding of the regression process.

We hypothesized that mothers of autistic children who regressed will try to receive diagnoses and interventions earlier than mothers with children who did not regress. We further hypothesized that mothers of children who regressed will be more likely to attempt to explain the processes underlying autism than mothers of children who did not regress. An additional hypothesis was that mothers whose children regressed later would be even more likely to search for explanations of the causes of autism, than mothers whose children regressed early. Furthermore, we were interested in comparing differences on demographic variables and medical history to determine if differences exist between children who regress and those who do not regress, and whether differences exist between the two subgroups, early and late regression.

METHOD

Subjects

Fifty-five mothers of all 56 children (43 boys and 13 girls—1 pair of twins), who were diagnosed with autism Disorder (American Psychiatric Association [APA], 1987, 1994) in the Haifa catchment area between 1989 and 1993 were contacted by phone by the first author. They were asked to volunteer for interviews. Thirteen mothers refused to participate and 2 could not be contacted. One child was excluded because of fragile X syndrome. Thirty-nine mothers participated and information about 40 children (29 boys, 11 girls) was collected including the pair of twin girls. The children of mothers who were not included in our study did not differ by diagnosis or place of treatment.

Procedure

This study employed a structured personal interview averaging 2 hours with each mother. Mothers were interviewed using a questionnaire designed for this study. Not all the results of the various medical investigations were included in the records we received from the different centers. Nor was developmental regression documented in a standardized manner in the medical records. Clinicians were also inconsistent in their recordings of mother's perceptions about their children's regression and their reactions to it. Therefore, medical records were examined for confirmation of the diagnosis only.

The questionnaire was composed of structured "closed" and "open" questions. The closed questions requested demographic information, immediate family members' medical histories; pre-, peri-, and postnatal period information; medical status and history; developmental milestones in gross motor skills (e.g., age of independent waking); fine motor skills (e.g., manipulation of small objects, puzzle); social skills (e.g., toilet training, imaginary play); language skills including number of words at 18 and 24 months; receptive language abilities (e.g., understanding simple commands) and the usage of nonverbal communication (e.g., gestures); language level at the time of the interview; and current school placement. In addition,

mothers were asked about any regression in each of the developmental areas. In the open questions, the mothers were asked their opinions of the diagnosis, their view of the diagnostic process and referrals for specific treatment. (See the Appendix for a sample of questions. A copy of the complete questionnaire may be obtained by mail.) Father's occupation was used for socioeconomic classification (Office of Population Census Surveys, 1971).

The mothers were unaware of the specific hypothesis underlying this study or the comparative analysis at the time of the interview. Three experienced interviewers, who were unaware of the previous history of the child, conducted the interviews. To establish reliability between interviewers prior to the study, the interviewers conducted simulation interviews, and for each interviewer two interviews were attended by a second interviewer. All of the mothers' responses were manually recorded and coded. Every interview was coded by a second interviewer and interrater coding reliability reached 95%.

Statistical Analysis

Regression was defined on the basis of mothers' responding "yes" or "no," and the mean age of regression (24 months) was employed to classify Early and Late regression. Chi-square analysis was used for comparison of perinatal complications. General linear model procedure was used to analyze the continuous data. The SAS package (1982) was employed for the analysis.

RESULTS

Of the 40 children in this study, 27% were girls. Nineteen (47.5%) of the 40 children were reported by their mothers to have regressed in at least one area. Of the 19 children, 16 boys (85%) and 3 girls (15%) were reported regressed ($\chi^2 = 2.489$, p = ns). As a result of the small number of girls in the regressed group, we were unable to perform further statistical analysis on gender. The children's mean age at time of interview was 6.89 years (SD = 1.55) for those who regressed and 7.26 years (SD = 1.78) for those who did not regress. Parents' age and children's birth order were similar for the regressed and the nonregressed group. Only 1 mother of a child who had not regressed reported mental illness in the past (depression); none of the fathers suffered from mental illness. The pair of twin girls in the study did not regress. One boy who did not regress had a sibling with autism. No relationship between socioeconomic status and reported regression was found.

Complications during the pregnancy were as prevalent among mothers of the regressed (n = 4) and of the nonregressed group (n = 4). One mother in each group had a cesarean section, and two in each group had an instrumental delivery. Epidural anesthesia was performed more frequently in the regression group (n = 6) than in the nonregressed group, χ^2 (N = 1) = 7.86, $p < 0.01$.

The mean Apgar scores that mothers had reported were very similar: nonregressed group 8.83 (SD = 0.61) versus 8.75 (SD = 0.85) in the regressed group. Reported birth weight was not significantly different

Mean age of regression was 24.05 months (SD = 9.43). Eleven children regressed before 24 months (early regression = ER), with a mean age of 17.18 months (SD = 4.06), and 8 children regressed after 24 months (late regression = LR) with a mean age of 33.5 months (SD = 5.39). Among the nonregressed group, early onset of autistic features (before 24 months) was reported for 15 children with a mean age of onset of 13.06 months (SD = 7.33), and late onset of autistic symptoms in 6 children with a mean age of 30.16 months (SD = 6.08). The mothers of this latter group did not report regression in any area.

The time when mothers reported experiencing first concerns regarding abnormal development was at age 17.95 months (SD = 10.46) for the nonregressed group, 17.36 months (SD = 4.82) for the ER group, and 33.5 (SD = 6.67) for the LR group. The time interval between problem recognition by the family and a definitive diagnosis was reported to be longer in children who had not regressed (12.23 months, SD = 7.68) than those for children who had regressed (6.84 months, SD = 8.46, F = 4.46, p = 0.04). However no significant difference was found between the ER and the LR groups (ER: 6.54 months, SD = 3.69; LR: 4.12 months, SD = 5.54).

Table I represents mothers' reports of regression in developmental areas according to ER and LR. Mothers reported regression in several areas including verbal and nonverbal skills, social and play abilities, with no significant difference report by mothers of the ER and the LR groups (Table I). Mothers did not report regression in motor abilities. Although mothers reported that children who regressed started to walk independently earlier (14.42, SD = 3.5) than those who did not regress (16.71, SD = 5.25), this difference did not reach significance. However according to mothers' reports, mean age of walking in the ER group (13.18 months) was earlier than that of the LR group (16.12 months) and that of the nonregressed early onset group (17.66 months) and this was statistically significant (F = 2.83; p = 0.05).

Medical evaluations as reported by the mothers were similarly distributed between the regressed and

Table I. Regression by Developmental Area According to Early and Late Regression

Area of regression	Early regression (n = 11) n	Late regression (n = 8) n
Loss of meaningful words	7	7
Receptive language	7	6
Non verbal communication	8	6
Eye contact	7	6
Social skills	3	3
Imaginary play	0	3
Nocturnal enuresis	1	1
Imitation	0	1
Decrease in body contact	3	1

nonregressed groups: EEG was performed in 17 and 15 children, respectively. The vast majority of mothers did not recall specific type of EEG performed. All results were reported as normal except for 1 child who had convulsions before the diagnosis of autism at the age of 6 months. Mothers reported that metabolic screening was done in 9 children who regressed and 15 children had not regressed and all were normal. Reported brain CT or MRI scans were performed on 8 children in each group, with no abnormal findings. Thirteen mothers whose children regressed and 16 whose children did not regress reported that genetic tests were performed and that results were normal.

At the time of the interview, the level of verbal communication, as reported by the mothers, was better in the regressed group, with no difference between the ER and the LR group (Table II).

When mothers of the nonregressed group were asked to state their child's main problem, nine answered autism or pervasive developmental disorder (PDD); an additional nine mothers reported communication/language problems, and three reported general developmental delay. Two mothers whose children showed ER and three of the LR group answered autism or PDD. Five in each group stated communication/language problems, and three of the ER group described general developmental delay. There was no significant difference in diagnostic reports of mothers whose children regressed and whose children did not regress between the groups.

Of the mothers of children who did not regress, seven (33%) had no explanations when asked about the reasons for their child's abnormal development, compared to only one mother in the regressed group. Mothers' explanations regarding the autistic features and the regression are presented in Table III.

When mothers were asked, given their accumulated experience, what they would have done differently in the past, more than half in each group stated that they would have asked for developmental assessment earlier, and for a more intensive treatment plan immediately following the diagnosis. Nine mothers whose children regressed expressed guilt feelings (regarding the pregnancy, delivery, or timing of house moving, or immigration) compared with three mothers of children who had not regressed (Table IV).

Table II. Level of Verbal Communication as Related to Regression Group.

Level [a]	Nonregressed (n = 21)	Regressed (n = 19)	Early regression (n = 11)	Late regression (n = 8)
1	3	4	3	1
2	14	7	3	4
3	4	8	5	3

[a] Level 1: No verbal communication, some uses gestures or augmentative communication; Level 2: Verbal communication, with augmentative communication; Level 3: Verbal communication.

DISCUSSION

Mechanisms underlying regression in a substantial proportion of children with autism have yet not been elucidated. Although the etiology of autism is also unclear, it would be reasonable to assume that a process of regression which involves such important loss of milestones will be accompanied by abnormal medical, familial, or social/environmental factors. Children with autism who regressed were reported to have lower verbal abilities, lower adaptive level, and lower IQs when compared with children with the same diagnosis who did not regress (Hoshino *et al.*, 1987). Kobayashi and Murata (1998) reported a significantly higher rate of epilepsy

Table III. Mother's Explanations of Their Children's Developmental Problems

	Nonregressed group (n = 21)	Regressed group (n = 19)
Pregnancy and delivery problems	5	5
Immunizations		1
Fever—neonatal period	1	
Death in the family		2
Convulsive disorder		1
Movement to new house or immigration	1	2
"Congenital brain problems"	7	7
No explanations	7	1

Table IV. Maternal View Pertaining to the Diagnosis and Treatment Course.

	Nonregressed group (n = 19)	Regressed group (n = 19)
Earlier evaluation and treatment and more intensive treatment	11	12
Change place of evaluation or treatment	6	3
Self guilt feelings	3	9
No changes	5	3

among the regressed group, while epileptiform EEG was also recorded more frequently in this group (Tuchman & Rapin, 1997). However, the role of subclinical epilepsy in the developmental regression of children with autism remains unclear (Rapin, 1998). In the present study we collected information from 39 mothers who were the only source of information (except the confirmation of the diagnosis). The intention of this study was not to explore the etiology of regression in autism but rather characterize familial as well as medical aspects related to this still unresolved phenomenon. The regression phenomenon is a catalyst for developmental assessment. Both parents and physicians are probably more alarmed in face of a regressive course. The fact that children regressed at an older age did not necessarily shorten the interval for evaluation. There was no difference in regression of developmental area between the LR and ER groups, with verbal and nonverbal communication being the most frequent in both groups. That regression in autism does not involve motor abilities was previously reported (Tuchman & Rapin, 1997), and was corroborated in this study. The age of walking onset represents an important gross motor milestone. Mothers of children in the ER group reported that they began walking even earlier when compared with the nonregressed group. In addition, no mother reported regression in other gross motor milestones. This suggests that regression may involve those brain areas responsible for autistic features but not to basic gross motor areas.

Parents' age, socioeconomic status, and family medical history were similar in the regressed and nonregressed groups. This was also found in the perinatal data, including the number of assisted deliveries. The meaning of the significantly more frequent epidural anesthesia performed during the labor of mothers whose children regressed is unclear. No differences were found in reports of the results of the medical workup between the children who regressed and those who had not regressed. Reports of medical workups were also unrevealing in those who regressed earlier and later in childhood. However, it should be noted that reports of frequency of investigation were similar among the regression and nonregression group. Mothers indicated their belief in the adequacy of medical investigation, regardless of their children's developmental course.

Mothers' reports of when they first became concerned about their children's autistic symptoms in this study were similar to the time recently reported (De Giacomo & Fombonne, 1998).

Unlike previous reports (Brown & Prelock, 1995, Kubayashi & Murata 1998), the current level of language ability (see Table II) was better in the children who regressed and there were no significant differences between children with early and late regression. This finding can only be suggestive, as we had to rely on maternal reports. It should be replicated by a comparison of clinical testing of language ability and development and parental report.

Childhood Disintegrative Disorder (CDD) is an uncommon condition in comparison with typical autism. Children with CDD reportedly have normal development for at least 2 years, before significant loss of acquired skills, occurring in language, social, motor, and play and the appearance of autistic traits (APA, 1994). This course, although it is more severe (usually in an older age), is similar to the regressive course seen in typical autism. Whether CDD constitutes a different entity is not clear (Burd, Ivey, Barth, & Kerbeshian, 1998). Children with CDD were found to be different from early onset (before 2 years) and late onset autism (after 2 years) (Short & Schopler, 1988; Volkmar & Cohen, 1989). Comparison of these three groups revealed that children with CDD had the lowest IQs and the late onset autistic group had the highest IQs. The children of the former group were also more likely to be mute and to be in residential placement. Except for age of symptom appearance, no differences were found between children with CDD and autism, in terms of familial factors, degree of autistic behavior, neurological factors, and courses (Militerni, Bravaccio, D'Antuono, & Palmina 1997).

When we evaluated mothers' reports, it appears that four children of the LR group could have been diagnosed CDD (APA, 1994). This would represent 10% of our study population. Previous reports point to a worse prognosis for this group. However, the CDD children were not different from the four children who also regressed later. Because of the small number of CDD and children who regressed late, no conclusions may be drawn.

Almost all of the mothers whose children regressed attributed specific causes to their child's autism, when compared to two thirds of mothers of

the nonregressed group. It is possible that regression is associated with different perceptions of the condition by parents. A proportion of mothers, especially in the regressed group, reported guilt feelings. It appears that these mothers feel guilty because they were confronted with a dramatic negative change in their child's behavior that they could not prevent. The multidisciplinary team who works with the family should address these guilt feelings.

When asked about the diagnosis, a larger proportion of mothers of both groups of children did not report their children as having autism although all were so informed. We may speculate that mothers who avoided using the term "Autism" are still in a stage of not accepting the diagnosis.

In conclusion, the results of this study do not indicate a significant difference between children who regress and who do not regress in mothers' reports of medical history, nor medical investigations. Similarly, perceptions of the diagnosis were not significantly different between mothers of children who regressed and those who did not. As this study is based on the total population of a cohort of children who were diagnosed with autism, it appears that the regressive course may be one of the typical phenomena characteristics of the autistic spectrum and may not represent a separate subclass of autism. Studies of larger populations of autistic children may shed more light on this issue in the future.

Regression is prevalent in about half of the children in this study and could not be either an exclusionary or inclusionary criterion for autism. These results suggest employment of additional investigative approaches, such as neuroimaging, prolonged sleep EEG, and auditory ERP, in an attempt to decipher the uniqueness of children with autism and developmental regression as compared to their nonregressed peers.

APPENDIX

Questionnaire for Mothers (Sample Questions)

A. Family and Demography

Mother: e.g., age, country of origin, occupation, medical history, developmental problems as a child.

B. Pregnancy and Delivery

Complications: e.g., bleeding, diabetes, hypertension.

Habits: e.g., smoking, alcohol.

Delivery: spontaneous, cesarean section, instrumental, epidural anesthesia.

Complications

C. Neonatal Period and First Year

Problems: e.g., breast-feeding, sleeping, feeding.

D. Medical History

Medical problems: neurological (e.g., epilepsy), nonneurological (e.g., asthma), sensory (e.g., vision, hearing).

Medical investigation: e.g., brain imaging, EEG, metabolic work-up, genetic consultation.

E. Developmental Report

Gross motor: age of independent walking. Have you noticed regression in gross motor abilities (e.g., running, climbing stairs or riding bicycle)?

Fine motor: Have you noticed any regression in fine motor abilities (e.g., playing with cubes or drawing)?

Social: e.g., In what age domestic mimicry, imaginary play, independent eating, and dressing had developed? Have you noticed any regression in these abilities?

Language: e.g., How many words did your child have at 18 and 24 months? Did he regress in this area (e.g., lost meaningful words)?

Nonverbal communication: e.g., What type of communication your child had used in the past? (e.g., pointing, hand leading, other gestures, augmentative). Have you noticed any regression in these abilities? At what age eye contact had developed and have you noticed any regression?

Level of verbal communication at the time of the interview: no words, less than 5 words, less than 50 words, sentences, normal?

Current school placement

F. Maternal Perceptions (Open Questions)

1. What in your opinion does your child suffer from?
2. When did you first start to have thoughts that your child is different?
3. In your opinion, what caused your child to develop differently?
4. What was the professional explanation you were given regarding your child's problem?
5. Was there any special event, or has your family experienced any kind of crises during the pregnancy, or the first years of your child's life?

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