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Parental reporting of regression in children with pervasive developmental disorders

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Abstract

In this paper we present the results of a systematic review of a series of cases reported by parents to have experienced some type of regression in their development in order to determine if there are significant differences between this subgroup of children and those without reported regression. Records were reviewed for all subjects assessed in a developmental disabilities clinic for approximately a 6-year period. Each case was grouped into one of four categories using a standardized method based on review of the record: 1) clear loss 2) possible loss 3) stagnation 4) no reported loss. Overall, parents reported some form of regression fairly frequently (18.8 % of the entire sample), however only 7.5% of cases were placed in either the clear or possible loss group, and 9.2% were placed in the stagnation group. There were significantly more subjects with either a clear or possible loss in the autism group ($x2=11.9$, $df=2$, $p<.02$) compared to the PDD and DD groups (11.8%, 5.5%, and 2.9% respectively). An analysis of the Autism Behavior Checklist scores and Vineland Adaptive Behavior composite standard scores covarying child age did not achieve statistical significance for the four loss groups. Therefore, parental reports of loss of skills were not reflected in a greater degree of severity as assessed by either instrument. However, the phenomenon of regression is clearly complex. In some instances the issue has more to do with developmental stagnation (failure to progress) rather than actual loss of skills. If a more stringent definition of regression is applied, where parental report of earlier developmental milestones supports the report of normal or near normal development, then rates of regression decrease. In future studies it will be important to devote considerable care to the identification of these more rigorously defined cases of regression.

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Parental Reporting of Regression in Children with Pervasive Developmental Disorders

Introduction

In his original paper Kanner (1943) emphasized the apparent congenital nature of autism. However, subsequent work (Kolvin, 1971; Volkmar, Stier, & Cohen, 1985; Short & Schopler, 1988; Volkmar, Cohen, Hoshino, Rende & Paul, 1988) made it clear that in a smaller number of cases, parents report normal development for 12 to 18 months before the development of typical autistic features. For example, in some cases early milestones such as sitting, walking, and first words are age appropriate, and then speech disappears only a few months after the child had begun using words (Kurita, 1985). In contrast to the rare syndrome of childhood disintegrative disorder (CDD) (Volkmar, Klin, Marans, & Cohen, 1997), where the child has progressed normally for several years, the 'later onset' autism cases usually have minimal speech skills at the time of the regression (10 or fewer words), and they generally seem to undergo a gradual process in which they fail to engage in communicative routines in which they participated in earlier. A complicating problem in this literature has been the reliance on parental report, which can be clouded by a number of factors (Lord, 1997; Volkmar et al., 1985). Other studies using early videotapes (Osterling & Dawson, 1994) have suggested that parents may not notice subtle abnormalities that antedate the time of their first concern. However, the issue of a possible subgroup of autism characterized by regression is of potential interest for various reasons including the recent concern that exposure to immunizations

might cause autism to develop (Wakefield et al., 1998). In this paper we present the results of a systematic review of a series of cases reported by parents to have experienced some type of regression in their development in order to determine if there are significant differences between this subgroup of children and those without reported regression.

Methods

Subjects

Records were reviewed for all subjects assessed in a developmental disabilities clinic for approximately a 6-year period. Parents routinely had completed a detailed developmental history questionnaire, which included information on the child's birth and medical history, early development and developmental milestones, and a series of questions about the age at which parents had first been concerned about the child's development. Parents were also asked, "Did the child seem to develop normally for a time and then lose skills? If yes, please describe". Cases were excluded from further consideration if the final diagnosis was of Rett's or childhood disintegrative disorder (where a regression is known to be part of the clinical picture) or if the relevant items on the questionnaire were not fully completed. The final sample of 573 subjects included 463 males and 110 females with a mean age of 7.9 years at the time of assessment. The subject's diagnosis was made according to DSM IV criteria (American Psychiatric Association, 1994), and included 237 cases (199 males and 38 females) with the final clinical diagnosis of autism, 199 cases (168 males and 31 females) with other PDDs (PDD-NOS and Asperger Disorder), and 137 cases with other developmental disabilities (mental retardation and/or specific language disorders) (96 males and 41 females). For analysis subjects were further grouped into three diagnostic categories (Autism, PDD, and DD).

Procedure

Each case was grouped into one of four categories using a standardized method based on review of the record: 1) clear loss group (parental report of clear loss of skills in any domain other than some nonspecific behavioral change), 2) possible loss group (either the parents were not sure, the loss was not dramatic or reflected a general parental concern rather than loss of specific skills), 3) stagnation group (parents reported a stagnation in development, e.g., with acquisition of one or two words but then no further word development), 4) no reported loss category (either no loss was reported, or the child's behavior was reported to have changed without loss of specific skills). Reliability was established for a subset (10%) of cases by the two authors with excellent overall agreement (Intraclass r=.89). Subjects were also noted to be delayed developmentally if any of four developmental milestones, again as reported by parents, were significantly delayed. The ages at or beyond which subjects were considered significantly delayed were smiling $\geq$ 6 months, sitting $\geq$ 9 months, talking $\geq$ 14 months, and walking $\geq$ 15 months. For each case, results of the Autism Behavior Checklist (Krug and Arick, 1980) and results of the Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cicchetti, 1984) were available; IQ/DQ scores were not analyzed given the large number of different tests used. All statistical analysis performed had significance levels set at .05 (two-tailed).

Results

The types of regression listed by parents are reported in Table 1. The most common category was loss of language skills with 65.6% of the parents reporting a loss in this area. Parents of children with autism reported regression more often than those in either the pervasive developmental disorder (PDD) or developmental disorder (DD) groups (22.4%, 16.1%, and 16.8% respectively), although this fell short of statistical significance ($x^2=3.3$, $df=2$, $p=.07$). Overall, parents reported some form of regression fairly frequently (18.8 % of the entire sample), however only 7.5% of cases were placed in either the clear or possible loss group, and 9.2% were placed in the stagnation group (see Table 1). There were significantly more subjects with either a clear or possible loss in the autism group ($x^2=11.9$, $df=2$, $p<.02$) compared to the PDD and DD groups (11.8%, 5.5%, and 2.9% respectively). In the autism group 11.8% were placed in the clear or possible loss categories, while 9.3% were placed in the stagnation group after review of the record (see Table 2).

Based on parental report only a total of 5 subjects (3 with Autism, 1 with PDD, 1 with DD) in the entire sample could be said to have clearly demonstrated a loss of skills; parents had reported normal early milestones in each of these cases prior to the report of regression. The remaining group of possible loss cases included thirty-eight cases; in this group parental reporting indicated some evidence of preexisting delay in over 50% of cases prior to the perceived regression.

An analysis of Vineland Adaptive Behavior composite standard scores covarying child age did not achieve statistical significance for the four loss groups. There were

also no significant differences between the loss groups with respect to Autism Behavior Checklist scores. Within the autism group there were significant differences among the loss groups on the age of the child when parents reported they were first concerned and age of the child when evaluated. The parents of subjects in the clear loss group were worried at an average age of 24.0 months, in the possible loss group at 20.1 months, in the stagnation group at 18.2 months, and in the no loss group at 12.3 months ($F=6.3$, $p<.001$). The loss and stagnation groups were significantly younger at the time of testing compared to the no loss group (5.2 and 5.3 years compared to 10.7 years for the no loss group $F=7.3$, $p<.001$) – likely reflecting the greater awareness of autism, regression, and the trend towards earlier diagnosis and assessment.

For the group as a whole there were significant negative correlations between the age when the child was first tested and Vineland composite standard scores ($r = -.48$, $p<.001$) and between the age when parents were first concerned and the age at which the child was evaluated ($r=-.56$, $p<.001$). Similarly, overall those subjects with an older sibling were on average significantly younger when their parents reported that they were first worried (15.3 months compared to 18.4 months, $F=7.7$, $p<.01$). There was not a significant relationship between the parents' level of education and occupation and the child's placement in the loss category groups in this study, or the age the child was tested.

Discussion

In the present sample, parents of children with autism were more likely than parents of children with other developmental disorders to report a loss of skills in the child's development. However, examination of the data suggested that the question of loss of skills is a complex one with some parents reporting a failure to gain (i.e., rather than an actual loss), other parents reporting a clear loss of skills (and one compatible with parental report of earlier developmental milestones being within normal limits), and a group reporting loss of skills that was questionable because the reported loss was minor or there was a lack of specificity in the parental report. However, no significant differences were noted for the autistic cases between the various loss groups with respect to scores on the Autism Behavior Checklist or on the Vineland Adaptive Behavior composite score, if the age was covaried. Therefore, parental reports of loss of skills were not reflected in a greater degree of severity as assessed by either instrument.

There was a significant difference in the Vineland Adaptive Behavior composite standard score if the age was not covaried due to the difference in the average age tested of the four loss groups, i.e., standard scores become lower in older subjects when rates of gain are lower than the rate of change in age. The study by Davidovitch, Glick, Holtzman, Tirosh, and Safir (2000), also showed older ages at the time of evaluation for those who did not regress which may contribute to their findings of lower achievement in that group compared to those who regressed.

In comparison, other studies (Brown & Prelock, 1995; Kubayashi & Murata 1998)

have reported different findings, e.g., lower language abilities in the group of children who regressed. It is possible in other studies that some of the children in the regression group could have been diagnosed with childhood disintegrative disorder (American Psychiatric Association, 1994); such cases are known to have worse outcomes (Volkmar et al., 1997).

Lotter (1966) reported that one-third of cases surveyed had an onset involving a setback in development, while Kurita (1985) reported that 37% of subjects with autism experienced a loss of words. The present study reports lower percentages of losses, however, this difference may be due to differences in the definition of "setback" or "loss". Lotter's definition was either a loss of some ability or failure to progress after a satisfactory beginning. In the present study, Lotter's second criterion was felt to imply developmental stagnation (failure to gain) rather than loss of skills. In addition, the present study eliminated those subjects with Rett's and childhood disintegrative disorder where a regression is inevitably observed.

In the autism group in this study subjects whose parents were worried later in life tended to have higher Vineland standard scores while those worried earlier had lower scores. This suggests the possibility that greater developmental difficulty contributed to earlier identification. The presence of an older sibling was also related to earlier age of recognition consistent with other reports (De Giacomo & Fombonne, 1998), i.e., parents who have prior experience of children's development may be more aware of abnormal development.

The study has various limitations. On the one hand it has the advantage of using various aspects of parental report and some independently derived measures of severity to address the issue of regression in autism and other developmental disorder. Parental reports of such regression are common in autism spectrum disorder as well as more strictly defined autism; these reports are also noted in children with developmental difficulties which are not part of the autism spectrum. Consistent with previous work various factors may act to promote or delay parental concern. Furthermore the phenomenon of regression itself is clearly complex. In some instances the issue has more to do with developmental stagnation (failure to progress) rather than actual loss of skills. If a more stringent definition of regression is applied (i.e., where parental report of earlier developmental milestones supports the report of normal or near normal development) rates of regression decrease. In future studies it will be important to devote considerable care to the identification of these more rigorously defined cases of regression. It is possible that studies which employ broader definitions of such regression may miss important aspects of clinical presentation and phenomenology. The relationship of regression in autism to the rare syndrome of childhood disintegrative disorder (CDD) also deserves further attention. At the present time, by definition, CDD is diagnosed only after age 2 when previous development has been normal. It is possible that at least some cases of regression in autism which occur before age 2 may represent the earlier manifestation of the same process or processes operating in CDD.

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Table 1

List of Most Common Losses and Categories

Category	Percentage	Reported Loss
Language Loss	65.6%	Word loss or stopped talking
Social Loss	50.0%	Lost eye contact or interest in social games or in other people
Behavioral Change	34.4%	Began stereotyped behavior or became irritable, anxious, tactilely defensive, or sensitive to noise and texture
Motor Loss	3.1%	Lost ability to walk or climb stairs

* A child could be included in more than one loss category

Table 2

Rates of Parental Report of Loss and Rates of Loss Based on Developmental Histories

	Autism	PDD	DD
Parental Report of Loss	22.4%	16.1%	16.8%
Clear or Possible Loss	11.8%	5.5%	2.9%

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