Infantile Autism with Speech Loss before the Age of Thirty Months

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Of 261 children with infantile autism, 97 (37.2%) showed a total loss of the articulation of meaningful words before 30 months of age (speech loss). Speech loss tended to appear more frequently in girls than boys. The median age of onset of speech loss was 18 months. Most of the lost words were those of ordinary baby talk. The autistic children with speech loss had developed a limited meaningful vocabulary earlier, but demonstrated more retarded mental development several years after their speech loss than those without such regression. The causation and the clinical importance of speech loss are discussed.


A certain proportion of children with infantile autism seem to show a peculiar pattern of early speech development, that is, some children with infantile autism lose totally their meaningful words before the age of 30 months. Furthermore, these children do not necessarily appear to exhibit more serious developmental problems before they lose their vocabulary. The British working party (Creak, 1961) cited this phenomenon, though the upper limit of age of onset was not given as a symptom in Point 7 of the 9 diagnostic points.

Every clinician participating in the treatment and the study of infantile autism is familiar with this phenomenon. Nevertheless, there have been few reports on this kind of phenomenon in infantile autism. Lotter (1966) reported in his epidemiological study that, among 32 autistic children, 10 children (31.3%) had an onset involving a developmental setback that included speech loss. However, he did not investigate the phenomenon of speech loss itself. Instead, he studied more general “loss of some ability” and “failure to progress after a satisfactory beginning.”

A detailed description of the phenomenon of speech loss still has not been published. It also remains unclear whether or not infantile autism with an early history of vocabulary loss is different, in its antecedent factors, behavioral symptoms, developmental characteristics, and prognosis from the other kind of infantile autism without vocabulary loss.

Therefore, I conducted a clinical investigation and a comparative study in order to clarify these points, employing a large number of children with infantile autism.

Subjects and Method

Subjects with infantile autism were selected from children who had first attended the Child Guidance Center, affiliated with the National Welfare Foundation for Disabled Children in Tokyo, from January 1975 to December 1980. This center had been a leading facility in Japan for diagnosing and evaluating children with infantile autism and other developmental disorders since its establishment in 1972. The children, accompanied by their parents, came from all districts of Japan, and one-third of them came from Tokyo or neighboring areas.

At this center, all children were examined during 4 days on Monday through Thursday by child psychiatrists, pediatricians, pediatric neurologists, orthopedic surgeons, ophthalmologists, and dentists. Psychologists, speech pathologists, and nurses also made clinical evaluations on attended children. As a rule, all children underwent electroencephalography, electrocardiography, cranial radiography, and other routine laboratory examinations. Detailed questionnaires on the development and medical histories of children were completed by parents.

A team of three child psychiatrists, including the author, reviewed all the clinical charts of the children. By mutual consensus, the diagnosis of infantile autism was made on 285 children, according to the Rutter’s (1978) criteria.

These children all had a history of delayed and deviated speech development, severely impaired social and interpersonal relationships, stereotypic and repetitive behaviors, and/or resistance to change, with the onset of symptoms before 30 months. They also
met the DSM-III diagnostic criteria of infantile autism.

In this study, speech loss was defined in its widest sense. The inclusion criteria were:

1. An episode occurred wherein the autistic children lost, before 30 months, the use of all meaningful words, which were expressed spontaneously by children in a similar situation, such as requesting food or complaining of hunger, addressing parents, greeting, replying when called by name, or telling names of familiar animals or objects. (The total loss of gestural expressions or imitative behaviors, before 30 months, in children who had not yet developed the verbalization of meaningful words, was considered to be the same kind of episode, because these abilities are usually closely related to the development of language.)

2. After the disappearance of vocabulary, children remained mute for a certain period, at least for approximately 6 months, regardless of whether or not they recovered their verbal ability.

The information on speech loss, such as age of onset and vocabulary before speech loss, was obtained from the precise description of early speech development in the clinical charts made by speech pathologists and psychologists. Additional information was obtained from the questionnaire on early speech development completed by parents.

Autistic children were classified into two groups: autistic children with speech loss and autistic children without speech loss, according to history. Autistic children, whose vocabulary decreased or changed but had not been totally lost before the age of 30 months, were included in the group of autistic children without speech loss.

Autistic children, younger than 30 months at the time of initial evaluation at the Child Guidance Center, were also classified into one of the two groups, if the presence or absence of speech loss before 30 months was confirmed at the time of the follow-up evaluation after 30 months of age.

In some autistic children, the loss of a word repeatedly alternated with the gain of another, with a short interval, during which the children appeared to be mute. This pattern was unclassifiable. Children who showed this pattern and children who lacked enough information to be classified into one of the two groups were not included among the subjects. There were 24 such children.

Finally, a total of 261 children with infantile autism were selected as the subjects of this study. Their ages at the time of initial evaluation ranged from 2 years 2 months to 14 years 10 months with a mean age of 5 years 2 months; included were 222 boys and 39 girls.

There were 97 children (37.2%) with infantile autism with speech loss and 164 children (62.8%) with infantile autism without speech loss. The mean age of 5 years 1 month in the former group did not differ significantly from that of 5 years 2 months in the latter group.

It was impossible to carry out standardized intelligence tests on many of the autistic children. Therefore, levels of mental development of the autistic children were classified into one of two grades according to development quotients (DQs), which were obtained from the Tsumori's Mental Development Scale (the Japanese equivalent of the Vineland Social Maturity Scale) at the time of initial evaluation. These two grades were: DQ of 60 and over, and DQ under 60.

Levels of meaningful words and self-help skills at the time of initial examination were evaluated with ordinal scales blind to the classification of the two groups of children. The levels of meaningful words were divided into two grades, that is, "meaningful words present" and "meaningful words absent." The levels of self-help skills, namely, bladder and bowel control, eating without assistance, and changing clothes without assistance, were divided into these three grades: "established," "incomplete," and "not established at all."

Experienced pediatric neurologists made global EEG judgments blindly to the classification of the two groups of children with infantile autism.

This and other information about family history, obstetrical records, past history, and development were all coded. These data were processed with release 8 of SPSS.

Results

Description of the Phenomenon of Speech Loss

In the group of autistic children with speech loss, 94 (96.9%) demonstrated an episode of loss of meaningful words. Another 3 boys (3.1%), who had not yet developed meaningful words by the time of initial evaluation, were included in the group of those with speech loss, according to the definition of speech loss, because they had shown definite episodes of complete loss of gestural expressions or imitative behaviors, such as waving bye-bye or greeting by bowing the head.

Before the onset of speech loss, 3 children (3.1%) could use two- to three-word sentences. The other 91 children (93.8%) could use only single words. In the majority of these children, the vocabulary before the onset of speech loss was of a few or several words, and these words were not used frequently. However, in 7 children (7.2%), the mothers reported that their children had spoken many words. The highest number reported was 30 words.

Most of the words that had been expressed and lost
consisted of typical infant speech. The most frequently used word was “man-ma” including its variant “uma,” which is typical Japanese baby talk for food. It was used by 36 children, when they were hungry or saw food. The next frequent word was “mama,” which was used by 15 children in addressing their mothers. (In Japan, “mama” is usually used by children in the same manner as in an English-speaking country.) Other frequent words were, for example, “wan-wan” (Japanese baby talk for a dog), “nyan-nyan” (Japanese baby talk for a cat), and “bye-bye.”

Two illustrative cases of infantile autism with speech loss were as follows (before the onset of speech loss, the first case showed fairly normal development, and the second one had already shown abnormalities).

Case 1. A girl aged 3 years 9 months, with an uneventful natal period, was born 2 weeks later than scheduled. Labor was induced. Her birth weight was 2900 g, and the neonatal period was uneventful. She passed early motor milestones as usual. She was not eager to be cuddled during her babyhood. She spoke her first word at the age of 1 year. She could express several words of baby talk, including “bye-bye,” to other people. She also played peek-a-boo. She pointed to a TV set or an electric bulb when their names were spoken. She imitated activities on the TV programs. But after 1 year 6 months, her verbal expression and interpersonal relationships gradually became poorer, without recognizable reason. At the age of 2 years, she was totally mute and demonstrated a lack of eye contact. She also had lost stranger anxiety. She strongly resisted change in the familiar route, when she went out. She liked to play solitarily, placing things side by side. She displayed an abnormal amount of aversion to what she ate.

She made slight progress afterward. But at the age of 3 years 9 months, she had not yet regained her former verbal skills. Instead, she could express only echolalia. She still showed a lack of eye contact and underresponsiveness and her interaction with other children also remained poor.

Case 2. The second case was a boy aged 6 years 3 months. His mother received medication (details are obscure) for the treatment of toxemia of pregnancy, during the last trimester. The delivery was protracted because of coiling of the umbilical cord, but he had no asphyxia. His birth weight was 3200 g. He passed early motor milestones rather early. He started walking at 11 months after an unusually brief period of crawling. He had shown hyperkinesis since he started walking. At the age of 1 year 6 months, he began to speak. He expressed “mama” and “papa” in addressing his mother and father. He could wave bye-bye. He liked his mother to read books to him. After the birth of his brother, when the patient was 1 year 8 months of age, he gradually lost words from his vocabulary and interest in books. He started to show social aloofness and stereotypic hand-flapping. At the age of 6 years 3 months, he remained mute and still showed hyperkinesis. He had not established self-help skills; however, his interpersonal relationships seemed to slightly improve.

Table 1 shows the tendency of speech loss to be more frequent in female autistic children (51.3%) than in male autistic children (34.7%). The boy-girl ratio was 7.6:1 in infantile autism without speech loss, and 3.9:1 in infantile autism with speech loss.
children; parental discord in 2 children; change of residence in 2 children; and fright in 1 child.

Parental ages at pregnancy, academic background of parents, order in sibs, and birth weight did not differ significantly between the two groups of children with infantile autism.

Identified prenatal and perinatal risk factors were: maternal physical disorders (for example, common cold, German measles), threatened abortion, x-ray irradiation in the abdominal and the lumbar regions, medication, toxemia of pregnancy, premature rupture of the membranes, preterm labor, postterm birth, breech delivery, vacuum extraction, forceps delivery, cesarean section, coiling of the umbilical cord, asphyxia, and profound jaundice.

The percentage of children who had at least one of these obstetrical risk factors did not differ significantly between the two groups (67% in the group with speech loss and 71.3% in the group without speech loss). The percentages of children who had multiple, that is, 2, 3, 4, 5, 6, and 7 obstetrical risk factors also did not show significant differences between the two groups. There was no single common obstetrical risk factor that appeared to have had causative or etiological importance for speech loss.

Table 3 shows that autistic children with speech loss developed meaningful words significantly earlier than those without speech loss. The distribution of ages of attainment of head control, sitting without support, and walking did not differ significantly between the two groups.

In 93 autistic children, both the age of onset of the first word and the age of onset of speech loss were identified. The longest interval between these two ages was 33 months. The intervals were within 6 months in 68 children (73.1%). In 23 of the 68 children, the intervals were less than 1 month.

Development after Speech Loss

The median of intervals between the age of onset of speech loss and age of initial evaluation was 38 months.

The percentages of children who attended nursery, kindergarten, grade school, and facilities for mentally retarded children were not different between the two groups.

By the time of the first evaluation with a mean age of 5 years 1 month, 31 children (32.0%) had regained meaningful words, and the others (68.0%) had developed no meaningful words at all since the onset of speech loss. In 29 of the 31 children, meaningful words reappeared more than 1 year after the age of onset of speech loss.

Table 4 shows that the rate of children with a meaningful vocabulary at the time of initial evaluation was significantly lower in autistic children with speech loss, in both sexes.

The rate of children who had the ability to change clothes, without assistance, was significantly lower in male autistic children with speech loss (9.1%) than in male autistic children without speech loss (24.8%) ($\chi^2 = 7.00, p < 0.01$). The rate did not differ significantly between the two groups of female autistic children.

Autistic children with speech loss tended to demonstrate, though not significantly, a lower level of establishment of bladder and bowel control, and of eating without assistance, in both sexes.

DQs were obtained on 89 autistic children with speech loss (mean age, 5 years), and on 143 autistic children without speech loss (mean age, 4 years 11 months) (the mean ages did not differ significantly between the two groups). There was no significant difference in DQ distribution between male and female children in each of the two groups. Table 5 shows that the rate of children with a DQ under 60 was significantly higher in autistic children with speech loss (78.7%) than in autistic children without speech loss (58.7%).
Levels of Mental Development between Autistic Children with Speech Loss and Autistic Children without Speech Loss

<table>
<thead>
<tr>
<th>Autistic Children</th>
<th>DQ Range</th>
<th>Total</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>Under 60</td>
<td>60 and over</td>
</tr>
<tr>
<td>With speech loss</td>
<td>70</td>
<td>19</td>
</tr>
<tr>
<td>Without speech loss</td>
<td>84</td>
<td>59</td>
</tr>
<tr>
<td>Total</td>
<td>154</td>
<td>78</td>
</tr>
</tbody>
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Corrected $\chi^2 = 8.87, p < 0.005$.

Induced sleep EEG, with triclofos sodium, was recorded on 95 autistic children with speech loss and on 161 autistic children without speech loss. In autistic children with speech loss, all the abnormalities were paroxysmal, and were found in 27 (28.4%). Of the autistic children without speech loss, 34 (21.1%) showed proxysmal EEG abnormalities. There was no significant difference in the frequency of paroxysmal EEG abnormalities, between the two groups and between male and female children in each of the two groups.

Discussion

There are two possible explanations for the result that the autistic children with speech loss showed a lower level of mental development than those without speech loss, at a median interval of 38 months after the onset of speech loss. One is that many of the autistic children who showed a loss of speech were those who had already presented a lower level of mental development before the onset of speech loss than those who did not. The other is that speech loss is a manifestation of some underlying regressive or deteriorating change, which may affect the subsequent early speech and mental development of autistic children.

Many of the autistic children with speech loss had already displayed various developmental problems before the onset of the loss of speech. These children seem to have already possessed some vulnerability of speech and mental development. Nevertheless, they do not necessarily appear to have manifested a lower level of mental development, before the onset of speech loss, than those without speech loss. They began to speak in meaningful words, significantly earlier and passed some early motor milestones, at about the same age as the autistic children without speech loss. The lost words were not different in their content from the usual baby talk of ordinary 1-2-year-old Japanese children, though the vocabularies before the onset of speech loss were generally small. In many of the autistic children with speech loss, autistic symptoms seem to have come out fully, after or along with the onset of speech loss.

Furthermore, some autistic children do not seem to have shown definite abnormalities, prior to the onset of a certain regressive change, such as a loss of speech. From the results of the clinical survey that was conducted by Rutter and Lockyer (1967), we can estimate that among 58 children who had infantile psychosis with onset before 30 months of age, 9 (15.5%) had the onset of psychosis after a period of "reasonably definite normal development." Harper and Williams (1975) described a period of normal progress, before the onset of the pathological process, in 94 (71.8%) of their 131 autistic children, with an age range from 3 to 22 years, though their criteria of classifying autistic children into two groups, the acquired group and the natal group, were different from those of the present study.

These results suggest that speech loss itself may have been a manifestation of some underlying regressive change. This explanation applies most reasonably to children who demonstrated fairly satisfactory development, before the onset of speech loss.

The level of mental development of autistic girls tends to be lower than that of boys. (Tsai et al., 1981); however, it was not the case in this study. Therefore, the tendency that speech loss was more frequent in girls than boys cannot explain why the autistic children with speech loss showed a lower level of mental development than those without it. At present, the reason for the observation that loss of speech tended to occur with higher frequency in girls remains obscure. The validity of this difference needs to be investigated further, with other samples of autistic children.

Of the prognosis of infantile autism, Eisenberg (1956) described that the ability to communicate verbally by the age of 5 years was an important prognosticating factor. He also suggested that autistic children who lost the ability to communicate by speech had poorer prognoses than those who did not. This point was supported by other researchers (Usuda and Koizumi, 1981; Wakabayashi, 1974).

Rutter et al. (1967) reported that autistic children with an IQ or a SQ under 60 had poorer prognoses than those with an IQ or a SQ over 60. Lotter (1974) also reported that a measure of speech was the best single predictor of outcome.

Hence, many autistic children with speech loss may have poorer prognoses than those without speech loss. But 32% of the children with infantile autism who had lost speech displayed a tendency of regaining communicative words. Therefore, only through a long-term follow-up study will we be able to evaluate the
ultimate difference between the two groups of autistic children, and the predictive validity of speech loss.

The phenomenon of speech loss seems to result from a combination of multiple causative and predisposing factors. In some autistic children, the vulnerability of speech loss may have been derived, in part, from the obstetrical risk factors, the high rate of which, in this study, was compatible with the results of previous research (Deykin and MacMahon, 1980; Finegan and Quarrington, 1979; Lobascher et al., 1970). Furthermore, psychosocial stresses appear to have played an important role in precipitating the loss of speech in some autistic children.

A high incidence of EEG abnormalities has been one of the most important bases of the view that a certain type of brain dysfunction plays an important role in the causation of autism (Gubbay et al., 1970; Kolvin et al., 1971). In this study, however, more than 70% of the autistic children with speech loss did not show EEG abnormalities. The role of neurophysiological impairments, as indicated by the presence of EEG abnormalities, in the appearance of speech loss is yet to be elucidated.

Infantile autism is a clinical syndrome that includes etiologically diverse conditions. The task now is to classify the syndrome into valid subgroups.

The characteristic course of early speech and mental development suggests that infantile autism featuring speech loss might be regarded as a clinical subtype of infantile autism. Further studies, however, are needed to evaluate this point. These should include comparative studies of behavioral symptoms and long-term prognoses between the two groups of autistic children.

References