

THE LOSS OF CRITERIA FOR MENTAL RETARDATION AND REGRESSION OF SYMPTOMS OF CHILDHOOD AUTISM DURING A FIVE-YEAR FOLLOW-UP – A CASE REPORT

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SUMMARY – It is usually held that most individuals with childhood autism have poor prognosis. According to our knowledge, a favorable outcome of a child with childhood autism is quite rare and inspires a number of controversies. A 4-year-old boy was diagnosed with childhood autism and mild mental retardation. Interviews and findings reported by other specialists were used in the diagnosis and follow-up that lasted for five years, along with parental counseling and therapy. After three years, regression of the symptoms of childhood autism and loss of the criteria for mental retardation were observed. The difference between the second and first examination was 30 and 24 IQ points. The boy attends regular school with individual approach. Childhood autism and mental retardation should not be viewed as static conditions. Early and intensive therapy is recommended. Some children that make good progress can attend regular school.

Key words: *Autistic disorder – psychology; Autistic disorder – diagnosis; Mental retardation – diagnosis; Intelligence; Cognition disorders; Child; Case report*

Introduction

It is a common belief that childhood autism refers to a diagnosis that implies poor prognosis¹. There are few outcome studies that have produced variable results. Assessing the outcome from different aspects (occupation/education, friendships, communication and independent living), Billstedt *et al.*² found a high percentage of poor outcome, i.e. very poor outcome in 57%, poor outcome in 21%, limited but acceptable outcome in 13%, fairly good outcome in 8% of cases, and none with good outcome. In addition, they report

on other associated problems in e.g., coexistence of epilepsy, severe self-injury, severe violence, hyperactivity, pubertal regression and presence of catatonia². Howlin *et al.* also report worrying data, with very poor outcome in 12%, poor outcome in 46%, fairly good outcome in 19%, good outcome in 10% and very good outcome in 12% of cases³. Eaves and Ho⁴ have recently reported on generally better outcomes connected with better opportunities currently available to autistic individuals. They found poor outcome in 46%, fairly good outcome in 32%, good outcome in 17% and very good outcome in 4% of cases⁴. Altogether, it seems that the majority of individuals with childhood autism will have life-long disturbances¹, i.e. many of them will be greatly dependent on others^{2,3}, however, we should not neglect recent opportunities available to individuals with autism⁴.

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The most important predictors of the adult outcome of childhood autism are intellectual level and communication abilities^{1,5}. However, it seems that interventions are also in relation with better outcome⁶.

We present a case of a 4-year-old boy diagnosed with childhood autism and mild mental retardation. During 5-year follow-up, the symptoms of childhood autism regressed along with loss of the criteria for mental retardation. We are aware that such a case study is not unusual; however, it seems quite intriguing for the overall dramatic improvement. Although some findings we refer to below describe such cases, they can only exceptionally be encountered in practice, e.g., in their more than 20-year follow-up, Nordin and Gillberg could only identify 12 out of 700 individuals as being 'cured' from childhood autism¹. According to literature reports, even today such a favorable outcome inspires a number of controversies and opens questions because it generally occurs due to unpredictable causes and only in some children⁷.

Case Report

First examination

A 4-year-old boy was examined by a child and adolescent psychiatrist using a standard procedure consisting of psychiatric interview and observation of the child in the presence of his parents. The parents described the child's behavior as 'wild', playing in isolation by himself, not responding to calls or direct speech; there was a serious risk for him to wander away to the woods or the field without letting the others know it. Psychological testing with the Brunet-Lesine scale showed the intelligence level in the category of mild mental retardation, but it was interpreted as an approximation because of the possibility that the low score was due to the low grade cooperation⁸.

The child is the younger of two children in the family. The family lives in the country, they run a farm, both parents completed elementary school. The child was born at term from normal pregnancy. Ultrasonography (US) showed high head volume, however, after 3 months it was within the normal range. The mother described normal motor development, first steps at 14 months, babbling in the breast-feeding period, at the age of 7 months he was able to repeat "mum" and "dad", but speech did not develop any further. At the time of initial examination, his speech was at the level

of a few functional phrases. The mother noticed first delays at the age of two years, especially in the area of speech development. There was no evidence of mental illness in the family.

During first examination the boy was hyperactive, looking as if he was 'in his own world', not socialized, he spoke only a few words, and was preoccupied with toys in his hands. One curious finding was that he could recognize letters of the alphabet. At home, he preferred watching TV, also occupied with various mechanical objects like TV remote control. The child and adolescent psychiatrist established the working diagnosis of 'childhood autism in observation'.

Additional diagnostic work-up and definitive diagnosis

After approximately three months, somatic examination did not show any decline from normal findings, including the results of metabolic testing, audiological testing and other findings (laboratory, electroencephalography (EEG), computerized tomography (CT) scan of the brain, and karyogram). Four months of the first examination, the patient underwent first follow-up psychological testing. Persistence of negativism was observed, however, satisfactory cooperation was achieved on testing. Global intellectual functioning was classified in the category of mild mental retardation: *Razvojni test Čturić* (RTČ, Čturić Developmental Test) IQ=58⁹ and *Sistematski psihološki pregled-3* (Systematic Psychological Examination-3) test IQ=64¹⁰, both results being comparable with the internationally known Stanford-Binet test. The estimated developmental age was 30-33 months (chronologic age, 51 months). Besides the presence of a narrow attention span, there were difficulties in directing movements both in macro- and micro-motor actions. Sporadically, he used phrases "going out", "taking stairs" and "juice", which even in this agrammatical form represented his intentions and wishes.

At the age of 4 years and 9 months, assessment by the Childhood Autism Rating Scale (CARS) and Psychoeducational Profile Revised (PEP-R) was made^{11,12}. These internationally used scales were applied by an experienced practitioner. The CARS total score was 30. The patient was evaluated by four points at the Listening Response and Level and Consistency of Intellectual response scales; by three points at the Imitation scale; and by two points at the Body Use,

Visual Response, Fear or Nervousness, Verbal Communication, Non-Verbal Communication, Activity Level and General Impressions scales. At other scales he was evaluated by one point, which was within the normal limits. In the profile of PEP-R developmental scale, total score was 74 and developmental age of 2.5 years. The scores on single scales were as follows: imitation 10, perception 12, fine motor skills 6, gross motor skills 8, eye-hand integration 7, cognitive performance 13, and verbal performance 18.

Definitive diagnosis was established when the boy was 4 years and 9 months old. The child's clinical picture was predominated by disturbances in social interactions. In the area of communication, there was evidence of delay in language development. A subspecialist in child and adolescent psychiatry, with years of experience in diagnosing autism, made the diagnosis according to the International Classification of Diseases (ICD-10): childhood autism (F 84.0) and mild mental retardation (F 70)¹³. The criteria from the American classification DSM-IV for childhood autism and mental retardation were also met¹⁴.

Therapy administered

On initial examination, it was recommended to start therapy before reaching definitive diagnosis. In the first year of therapy, during autumn and winter, the boy attended individual sessions held by a special teacher, 2.5 hours *per week*. Progress was evident at follow-up visits during this period. In springtime, therapy was continued, once a week for 2 hours. At the next follow-up visit, the child showed an increase of interests, could read fluently, and was socialized in his family surroundings in his own way. Then a half-year break followed. In the second year of therapy, the boy started daily treatment in a kindergarten for children with special needs. After that, in the third year of follow-up, the boy attended regular public preschool program, twice a week for two hours. At the beginning of preschool treatment, he completed his individual special teacher therapy. Clinically, there was evidence of improvement; the speech/language performance was more focused on interaction. In the fourth year of follow-up, in autumn, the boy started attending a local elementary school, regular program only minimally modified with the so-called 'individualized approach' in teaching methods. The first year

of education ended with excellent marks. In the fifth year of follow-up, the boy was in the second-grade school program.

Follow-up testing and loss of criteria for the diagnosis of mental retardation and regression of autism symptoms

Second testing by the same psychologist was performed two and a half years after the first psychological testing, in the third year of follow-up, at the time the boy attended preschool program, at age of 6 years and 8 months. The boy quickly accepted the individual test situation and continued to cooperate, showing adequate interest in the test material, aware of his success, giving 'high-five' by slapping the examiner's palm, finally commenting that all 'went well'. Global intellectual functioning was classified at wider borders of the normal range for that age, in the category of low average range, with REVISK IQ=88 (verbal IQ=84, performance IQ=93)¹⁵(a test comparable to the internationally known Wechsler Intelligence Scale for Children). Performance or the nonverbal aspect was surprisingly classified within the average range, and verbal aspect within the lower average range. Even more interesting was distribution of the results in single functions, ranging from the high average in attention span (short-term memory) to the series of functions in the average range (information fund, visual detection, numerical reasoning, object assembly, visual analysis in Koch's subtest), then in the low average range (vocabulary) and in the borderline range in the function of perceptive-motor speed (the measure of attention persistence). Finally, the function from the social intelligence spectrum was classified in the range of severe mental retardation, i.e. extremely immature reasoning based on previous experience. The results represented enormous progress from previous intellectual functioning.

So, in the third year of follow-up the child was still attending preschool program. Upon psychological verification of the absence of mental retardation, the psychiatric report clearly indicated the absence of criteria for mental retardation. Clinically, significant improvement was evident. According to the latest psychological report, regular educational program at local public school was recommended, along with an 'individualized approach' in teaching methods. Further stimulative treatment by a special educator was also recommended.

In the fifth year of follow-up, reassessment was done by the same examiner using CARS and PEP-R scales. In CARS, total score was 19. The patient was evaluated with 1.5 points in the scales: Relating to People, Imitation, Emotional Response, Verbal Communication, Non-Verbal Communication, Activity Level, Level and Consistency of Intellectual Response, and General Impressions. All other scales were scored with one point. On the PEP-R developmental scale, total score reached maximum of 131 and developmental age of 6.5 years, suggesting the boy was functioning at the borderline intellectual level, an estimate made from the developmental age above. At this point of the child's development, the PEP-R scale was no longer sensitive to further progress and maturation, i.e. estimation of developmental age was no longer accurate, it could be higher but the scale would not recognize it. In this fifth year of follow-up, all findings showed improvement. He attended second-grade class at the local public school, with 'individualized approach' used within the scope of the regular educational program.

The last, third psychological follow-up testing at age of 8 years and 10 months confirmed stabilization of cognitive development. REVISK IQ=89 (verbal IQ=89, performance IQ=89), and total score confirmed global intellectual functioning at the low average level¹⁵. This time the conformation of the two aspects was obvious, which is commonly expected in the process of maturation. Yet, considering the distribution of single functions, disproportion was still present in their development, from the high average range (visual analysis in Koch's test, short-term memory), through average range (information fund, visual detection, object assembly, vocabulary) to the low average range (numerical reasoning, conceptual thinking) and borderline range (perceptive-motor speed, interpretation of new social situations), and finally reasoning based on life experience in the range of mental retardation.

Clinically, the patient was described as more cooperative, calmer, communicative, answering the examiner's questions properly, sometimes in agrammatical speech patterns. He had an urge to tell various stories; he filled series of notebooks with theatrical plays. He finished the first year of school with excellent marks and was able to tell the names of all pupils in his class and of his friends. While travelling to the hospital for

examination, he was fascinated by counting electricity poles, he could recite poems by heart, while playing alone he would largely comment aloud; he learned the multiplication table and was especially fascinated by division. Considering the great improvement/fading of the symptoms of autism, the diagnosis of childhood autism (F 84) was abandoned and evaluation of a residual category, which is another form of pervasive developmental disorder according to ICD-10 was established (F 84.8)¹³.

Discussion

Diagnostic procedure and therapy

Diagnostic process involved multidisciplinary experts, in line with recommendations for the diagnosis at earliest stages^{16,17}. The process of diagnosing could be extended to a longer period in order to establish definitive diagnosis, i.e. the diagnosis needs to be constantly reviewed. The classification diagnosis of autism was supported by a multidimensional approach, the use of CARS and PEP-R scales that indicated a milder form of autism^{11,12}. This can be important in future planning of therapy, which will not only depend on the activities to minimize inappropriate behavioral patterns, but will focus on developing healthier mechanisms and abilities. These findings do not differ significantly from other findings in international reports which suggest the first specific diagnosis to be reached relatively late¹⁷. The pediatrician or primary care physician should definitely take a more active role in screening for disorders that can be recognized in the first and second year of life¹⁷.

In our patient, individual therapy was recommended immediately after the first examination of the child, although the diagnosis was not confirmed, which is by no means rare approach in childhood and adolescence. In our case, the therapy applied showed favorable effects within a short period of time. The parents' cooperation was especially important to us. The therapy administered in the first year could be characterized as low-intensity outpatient therapy and as high-intensity in the second year, in accordance with the experience reported elsewhere¹⁸. In the third year, the specific stimulative therapy by special teacher was no longer applied and the child attended a preschool program instead, which most likely had

a favorable structural impact. This structural impact seemed to have continued in the fourth and fifth year of follow-up, while attending regular school. In their report, Harris and Handleman emphasize the importance of early intensive therapy¹⁹. Indeed, the children receiving such therapy were more likely to achieve successful inclusion in regular school program.

The loss of the criteria for mental retardation and regression of the symptoms of childhood autism

The most intriguing question we encountered in this case study referred to the remarkably explicit general improvement of the clinical picture, manifested as regression of the symptoms of childhood autism, increased level of functioning and loss of the criteria for the diagnosis of mental retardation, which is rather unusual^{1,5}.

At initial examination, the boy was much like an authentic 'feral child', similar to the children with childhood autism described or the well known story of Victor of Aveyron²⁰. The autistic symptoms gradually declined. In addition to clinical improvement, the patient was objectively evaluated by CARS and PEP-R^{11,12}. As for cognitive testing, the difference in global IQ between the first and second testing was 30 points (RTČ-REVISK) and 24 points (SPP-3 – REVISK), depending on the technique employed, which is highly significant according to the study that emphasizes the role of difference greater than 15 points of the scale¹⁸. It was also consistent with some reports on increasing IQ in some children with childhood autism over time^{16,18,19}. We are aware that sometimes a rise in IQ scores need not necessarily mean overall improvement of functioning²¹. In our case, the increase in IQ score occurred collaterally with regression of autistic symptoms, which is consistent with a previous study of the relation between IQ and autistic symptoms⁷. We hypothesized that the increase in IQ may have been due to a combination of maturation (neurobiological) and environmental (therapy) factors, which is consistent with some hypotheses proposed in the literature^{6,18,19}.

The present report may be criticized for not making use of the internationally acknowledged intelligence tests and for using different tests during follow-up. However, the tests employed have been commonly used in our social culture, as seen from a series of studies. Also, the clinical psychologist having admin-

istered the tests has collected more than twenty-year experience in testing, which reinforced the IQ findings recorded in our patient. Furthermore, literature reports reveal that it is by no means unusual to use different tests, adjusted to the subjects' age^{16,21}. Another possible objection to the present report could refer to the fact that special structured interviews (ADI-R and ADOS), commonly used in the diagnosis of childhood autism, were not employed^{22,23}. However, we consider our arguments were strong enough to make the diagnosis. We used clinical interview, which remains the 'gold standard' in the diagnosis of childhood autism²⁴, along with ICD-10 and DSM-IV, which has stationary use in our practice^{13,14}, and the team of experts has more than 10 years of practice in diagnosing childhood autism. Finally, the child was 4 years and 9 months old when the diagnosis of childhood autism was established, i.e. the age when the clinical picture is clearly expressed¹⁷.

Conclusion

Although literature reports indicate poor overall outcome, the case presented points to the need of dynamic assessment of diagnostic criteria over time in each individual case. Indeed, the improvement of clinical picture is possible, and so is the loss of the criteria for each diagnosis. The importance of continuous therapy is stressed, and it should be recommended at the very first examination, which will increase the probability of attending a regular school program.

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Sažetak

GUBITAK KRITERIJA MENTALNE RETARDACIJE I SMANJENJE SIMPTOMA AUTIZMA U DJETINJSTVU TIJEKOM PETOGODIŠNJEG PRAĆENJA – PRIKAZ SLUČAJA

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Općenito se smatra da je autizam u djetinjstvu u većine osoba lošije prognoze. Pozitivan ishod djeteta s autizmom je prema našim saznanjima prilično rijedak i pobuđuje niz proturječja. Četvorogodišnjem dječaku je bio dijagnosticiran autizam i blaža mentalna retardacija. U dijagnostici i praćenju koje je trajalo oko pet godina primijenili smo klinički psihijatrijski intervju kao i nalaze drugih stručnjaka. Primijenjeno je savjetovanje roditelja i terapija. Tri godine od prvoga pregleda kod dječaka je zabilježena regresija simptoma autizma uz gubitak kriterija za mentalnu retardaciju. Razlika globalnog IQ između drugog i prvog ispitivanja ovisno o tehnici bila je 30 i 24 boda. Dječak danas polazi osnovnu školu po redovnom programu uz individualizirani pristup. Mentalnu retardaciju i autizam u djetinjstvu ne treba shvaćati statično. Preporuča se rana intenzivna terapija. Pojedina djeca koja dobro napreduju mogu polaziti redovnu školu.

Ključne riječi: Autistični poremećaj – psihologija; Autistični poremećaj – dijagnostika; Mentalna retardacija – dijagnostika; Inteligencija; Spoznajni poremećaji; Dijete; Prikaz slučaja