ORIGINAL ARTICLE

The natural history of infantile autism in a tertiary pediatric public hospital: developments with global developmental delays with full and partial regression

A história natural do autismo infantil em um hospital público pediátrico terciário: evoluções com atrasos globais do desenvolvimento, com regressão parcial e plena

Walter Camargos Jr[1], Thelma Ribeiro Noce[2]

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ABSTRACT

Autism is a behavioral severe disorder characterized by a start within 36 months of age, with symptoms in the areas of interpersonal interaction, communication, and behavior. It presents three forms of evolution: always with delay in psychic development, loss of some acquired skill on the basis of a delay in development (partial regression), and full regression. **Objective:** to research the frequency of each form of evolution. **Methods:** longitudinal study in a tertiary pediatric public hospital in an outpatient Developmental Disorders Clinic, during 20 months, with the first appointment within 48 months of age. The DSM-IV and the CARS and DSM-IV scales were used as diagnostic criteria; questions were asked about regressions and delays in development or not in the initial development of autism. **Results:** of the 86 children with clinical signs and positive scales for autism, 74.4% always showed delays in development, 17.4% showed a history of delays in development with some degree of regression, and only 8.1% presented a history of full regression. **Conclusion:** all children with a history of permanent delay in the milestones of psychic development should have autism as the diagnostic hypothesis.

Key words: Autistic Disorder; Prevalence; Clinical Evolution.

[1] MD, Psychiatrist. Master degree in Health Sciences. João Paulo II Children's Hospital from the Hospital Foundation of the State of Minas Gerais (FHEMIG). Belo Horizonte, MG – Brazil.
[2] MD, Neurologist. Master degree in Medical Science in the Neurology Area. João Paulo II Children's Hospital from FHEMIG. Belo Horizonte, MG – Brazil.

RESUMO

O autismo é um transtorno comportamental, grave, caracterizado pelo início até 36 meses, com sintomas nas áreas de interação interpessoal, comunicação e comportamento e apresenta três formas de evolução: sempre com atraso no desenvolvimento psíquico, perda de alguma habilidade adquirida em base de atraso no desenvolvimento (regressão parcial) e regressão plena. ***Objetivo:*** *pesquisar a frequência de cada forma de evolução.* ***Métodos:*** *estudo longitudinal em hospital público pediátrico terciário, durante 20 meses, em Ambulatório de Transtornos do Desenvolvimento, com primeira consulta até 48 meses. O DSM-IV foi utilizado como critério diagnóstico assim como as escalas do CARS e do próprio DSM-IV e foram feitas perguntas sobre regressões e atrasos no desenvolvimento, ou não, na evolução inicial do autismo.* ***Resultado:*** *das 86 crianças com sinais clínicos e escalas positivas para autismo, 74,4% sempre tiveram atrasos no desenvolvimento, 17,4% tinham histórico de atrasos no desenvolvimento com algum grau de regressão e somente 8,1% apresentaram histórico de regressão plena.* ***Conclusão:*** *todas as crianças que apresentam histórico de permanentes atrasos nos marcos do desenvolvimento psíquico devem ter o autismo como hipótese diagnóstica.*

Palavras-chave: *Transtorno Autístico; Prevalência; Evolução Clínica.*

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Institution:
João Paulo II Children's Hospital – FHEMIG
Belo Horizonte, MG – Brazil

Corresponding Author:
Walter Camargos Jr
E-mail: waltercamargos@uaivip.com.br

INTRODUCTION

The term infantile autism[1] is today known as the result of the association between autism itself with mental retardation (MR), to some degree, also named as low-functioning autism spectrum disorder (LFASD) versus the high-functioning autism spectrum disorder when no MR is present as a comorbidity. Epidemiological studies show that the prevalence for the entire spectrum of autism is between 0.9%[2] and 1.5%[3] with 70% of this population also presenting MR.[4] Infantile autism affects more men than women (4M: 1F), is considered a behavioral syndrome, starts at 36 months of age[5], cause qualitative and quantitative losses in social interaction and communication, and present restricted, repetitive, and stereotyped behaviors.

Symptoms in the area of interpersonal interaction explain the name "autism", which manifests as: behaviors of less interest for people than to objects, greater interest in the internal/imaginary world than in the external world (people); the harm is more evident in children of the same age and unknown children but can also occur with relatives; less frequency looking at people, sometimes evident already during breastfeeding and care in the first months of life; little response/reaction when called by the name generating the initial suspicion of temporary deafness even when the child demonstrates listening songs and advertisements. They might also lack in demonstrating emotions in the absence of parents or their return, and may accompany or go to the lap of strangers without strangeness; seek for little comfort when falling and getting hurt; take people by the hand to get/do what they want (use people as a tool), etc.

Communication is impaired in the verbal and nonverbal dimension. There is less looking at the mother, family, and caregivers when directing their speech and more interest and responses in other sounds such as music; delay in speaking words and phrases; delay in the use of the pronoun "I", which is replaced by the third person pronoun; little or no use of communicative speech when aiming to interact, ask, or talk, but can sing – they speak without dialogue; echolalia (repetition of words, phrases heard, dialogues from videos); continuous or episodic sound making that does not have the means of interaction; verbal stereotypes (expression of sounds or words, automatically and without apparent purpose), among other signs.

There is absence or delay in the proto-imperative (point whatever) and proto-declarative (point to show) communication in the non-verbal communication as well as in the use of social signs (hand fan for farewell, thumb upward – "cool", clapping, etc.); least demonstration of facial mime in moments of interaction ("my son/daughter is very serious"); least demonstration of mime of satisfaction after feedings; discordant mime between verbalization and acts, such as a hug/kiss without affective mime expected; stereotyped body postures; clumsiness, or extravagant walk; among other signs.

Regarding behaviors, the following can be identified: hyperactivity or unusual passivity; tantrums, nervousness and laughter without a specific reason; hypersensitivity with signs of displeasure to abrupt sounds such as horns, fireworks, blender, and vacuum cleaner, etc.; self-aggression such as biting own hand and hitting own head on the floor; motor stereotypes (the most common is flapping arms like wings – "flapping"); intense pleasure when seeing rolling and bright objects; line up objects according to color and size; carry and cling to objects that do not have any apparent function such as strings, papers, chopsticks; non-functional use of toys (instead of playing with a car they keep turning its wheel); specific and strange interest in shapes, smells, texture to the touch, taste (e.g. lemon, pepper), which leads these children to catch, sniff, or lick everything; they play in a delayed way for their age (e.g.: seven-year-old boy who stays all day playing with filling and empting a sand bucket); decreased sense of danger; adherence to task routines; sleep alterations; absence of appeals to be fed or its opposite; among other behaviors.

When looking at these signs through the bias of development, the delay landmarks of the psychic development are evident such as, for example, five years old children show responses similar to those from babies, both in the quality and quantity of responses in all three affected dimensions (interaction, communication, and behavior).

It is important to remember that the deambulation (landmark of motor development) is almost a prerequisite of psychic development and its lacking prevents the acquisition and/or evidence of several psychic abilities, some of which are researched in the diagnostic scales, both in the Diagnostic and Statistical Manual of Mental Disorders IV[6] (DSM-IV) (items A. 1 b. A 2 d., and A. 3.a) and the Childhood Autism Rating Scale (CARS)[7] (items 4, 5, and 13). The lack of walking skills harms the reliability in the scales in addition to the indication of neurological disorders.

The natural history of the installation of infantile autism occurs in three forms[8]: always lagging behind in development and with progressive improvement; starting with delays in development and presenting partial regression (loss of some skills), and with normal development and full regression (loss of many skills of development already achieved, such as interaction and communication) without/with autism clinical installation after the uncontested status of organic base such as AVC, TCE, epileticus status, meningitis, and etc. The most publicized form by the lay media [9.10] is the full regression, and this can harm the chances for an early diagnosis. Most of the citations found in the literature on this subject are nonspecific, without discrimination of partial or complete regression. Regressions were found in 23.9% of cases by Parr et al.[11]; between 20 and 33% by Chawarska et al.[12]; 19% by Fombonne[13]; and 22% by Meilleur and Fombonne[14]. There is one single author[15] who reports that more than 50% of cases of regression always had delays in development.

The aim of this study is to evaluate, separately, what is the actual proportion of occurrence of these three forms of installation of infantile autism. The authors' hypothesis is that the form that occurs always associated with delays in development has the highest prevalence.

METHODOLOGY

This was a cross-sectional study conducted in a public pediatric hospital, tertiary, and recognized by the Ministry of Education and Culture as a teaching hospital.

The children assisted in elective consultations in the Complex Disorders Clinic of Child Development from João Paulo II Children's Hospital (HIJP-II)/Hospital Foundation of the State of Minas Gerais (FHEMIG) were evaluated between November of 2009 and June of 2011, at ages up to 47 months, scheduled by spontaneous demand.

The inclusion criteria were: presenting clinical picture of infantile autism; diagnostic confirmation by the scales from the Diagnostic and Statistical Manual of Mental Disorders IV[6] (DSM-IV) and the Childhood Autism Rating Scale (CARS)[7] at 36 months of age, parents/guardians agreeing and signing the Volunteer Informed Consent to participate in the study.

The exclusion criteria were: parents/guardians who did not have the capacity to understand the formulated questions; those who have development evolved enough to not fit the diagnose of positive case according to the DSM-IV and CARS scales up to 47 months of age; clinical pictures with indisputable evidence of neuro-organic causes (that could generate significant delays in development); those who did not deambulate, and those under 24 months of age.

The history of the installation of infantile autism was researched in parallel to the diagnosis through the use of a brief questionnaire.

INSTRUMENTS

The instruments used were the two diagnostic scales, DSM-IV (6) and the CARS[7], and a brief questioning of parents/guardians about the evolutionary characteristics of autism in the first 36 months of the child's life. The mark of 36 months of age is given as one of the DSM-IV criteria for the diagnosis in which the child must already have presented qualitative and quantitative losses that are consistent with infantile autism. Conversely, families of children older than 47 months of age commonly no longer have the accurate information about the process for the installation of the clinical picture, which is the goal of this research.

Parents were asked to report in detail how the installation of the picture of autism had occurred focusing on the following three possibilities:

a. if the development had always been late;
b. if there was any normal skill that was lost such as talking (talking with intent to communicate); use of intentional social signals – waving, raising the thumb up; pointing to communicate the desire to show, as a way to share, etc.; however, in variety and quantity of course smaller than the normal. Echolalia (repetition of words and phrases), repetition of numbers and letters were not considered a normal prior skill;
c. if there were normal development with full regression, and, if occurred, if there was an evident neuro-biological picture preceding the development regression.

A neuro-pediatric clinical evaluation was performed to assure possible evidence of organic base events that could explain the delay in development better than autism itself.

ETHICAL ASPECTS

The research was approved by the Ethics Committee from the Hospital Foundation of Minas Gerais

State, registered in the SISNEP, and all parents/guardians signed a Volunteer Informed Consent form.

RESULTS

During the 20 months of study, 106 patients (84M and 22F) were evaluated. Of this group, 20 patients were excluded, resulting in 79 males and 7 females with the median age of 36 months of age; all patients presented the clinical criteria of infantile autism confirmed by the DSM-IV and CARS diagnostic scales.

The clinical characteristics of the excluded group were: four (2M: 2F) did not present the clinical picture of infantile autism; four boys have evolved to the point of no longer being positive on the scales at 48 months of age; three boys were younger than 24 months; two boys and one girl could not walk; one boy had the West syndrome; two girls had microcephaly; one had the diagnostic hypothesis of Rett syndrome and the other of inborn errors of metabolism; and there was one boy who was excluded because of questions about the presence or absence of the late development causative disorder.

In relation to the three evolutionary forms, 74.4% (53M: 11F) always had delays in development, 17.4% (12M: 3F) had partial regression in a deficient basis of development, and only 8.1% (6M: 1F) had a history of full regression (Table 1).

Table 1 - Comparison between the three evolutive forms

	Male	%	Female	%	Total	%
Always with delay	53	61,6	11	14	64	74,4
Partial regression	12	14	3	2,3	15	17,4
Partial regression	6	7	1	1,2	7	8,1
	71	82,6	15	17,4	86	100

On a more pragmatic vision, 91.8% were found (65M: 14F) with a history of delays in development (sum of the result of the group who always showed delays in development with the group of partial regression) and only 8.1% (6M: 1F) had full regression history.

A new fact was detected during the research in the group of those who have evolved with partial regression, which were the major environmental changes in the chronologically concomitant history preceding the partial regression installation, such as moving to a new house, new babysitter, birth of a brother, death of people whom the child lived intensely close, father who went to work in another city and was not present in the same frequency as before, etc. This information was found for 6 out of the 15 patients in this group (five in 12 boys and 1 in 3 girls); this question was not researched in the first examination.

DISCUSSION

The clinical picture of infantile autism in children over five years old is already well recognized by medical professionals; the recognition of its natural history is also important for an early diagnosis. This is the first study that surveyed separately the three possible types of the initial evolution of infantile autism.

The comparison between the overall frequency of regressive events, which is 25.5% (17.4% added to 8.1%) of the sample with those in the literature shows similar results, between 20 and 33%, and close to those reported by Parr[11] - 23.9%.

Of more practical importance is the fact that 91.8% of the researched population and with IA evolves with serious psychic delays (to interact; communicate; play, etc.) until the diagnosis is established. This information is important to public health in the country because with the focus on psychic development delays, the early diagnosis would be possible and, if associated with appropriate treatments and specialized fast therapy, there would be increased chances of clinical improvement and, consequently, psychological, financial, and social lower costs, both for families and the State, notably for the population of low income.[16]

This result (8.1%) goes against the speech from a large number of families and the media[9,10] assuming that the installation of IA mostly occurs with total regression.

Another point of relevance in the discussion is that all health professionals, doctors or not, might suspect this disorder based on the knowledge of the milestones of physical development. This procedure can also be aided by scales of suspicion[17] or diagnoses that are closer to Pediatrics than to the area of child Psychiatry, therefore there is no need to have training in childhood Psychiatry to be able to suspect and even formulate a diagnostic hypothesis.

The young age in the studied children during the course of the survey of information about the installation of the picture certainly favored the precision in the results.

These results also show that the hypothesis initially raised by the authors was correct; the highest prevalence of natural history of infantile autism associated with delays in development.

The novelty of finding about the concomitant evolution of partial regression and important modifications in the environment is perfectly understandable because difficulties in adapting to changes in routines are part of the clinical picture and are covered in specific items in the DSM-IV (a. 3. b) and CARS (item No. 6) scales, often generating disturbances/behavioral changes evident in this population. Because this was not part of the initial objectives of the research, there are medical charts where this information is not clearly written, which mean that this number may be higher.

CONCLUSION

The most important result in the research is the need to consider the hypothesis that infantile autism could be suspected whenever the patient presents a history of delays in development in the psychic landmarks of inter-social interaction, communication, and play. This information is also relevant in the implementation of public health policies because it is known that the treatment of a person with autism has specific characteristics and needs that are much more complex than the treatment of the population affected only by mental retardation.[16]

More complex studies are needed to verify and supplement the results of this study.

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