

CEREBROVASCULAR DISEASE IN CHILDHOOD

Neuropsychological investigation of 14 cases

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ABSTRACT - There are few studies about the development of a child after a cerebrovascular accident (CVA), and they usually describe problems such as diminishing in intellectual capacities, difficulties in linguistic and visual-motor skills, as well as in spatial organization and integration. In this study, there were 28 children participating, being 14 placed in the experimental group (EG) after clinical diagnosis and ischemic CVA imaging, and other 14 children without past history of CVA, who formed the control group (CG). The neuropsychological research protocol included an intelligence test, a visual-motor coordination test, human figure drawing, a cortical functions battery and the medical records of the children from the EG. The analysis of the results of this study revealed that the best performances after the CVA are related to the shortest time of functional recovery; CG presented better performances than EG in all the instruments used, in cognitive, perceptual and motor skills. It has been noticed that CVA may lead to intellectual reduction in case of a recurrence of the vascular insult.

KEY WORDS: cerebrovascular disease, childhood, neuropsychological investigation.

Doença cerebrovascular na infância: investigação neuropsicológica em 14 casos

RESUMO - Nos poucos estudos acerca do desenvolvimento da criança após o acidente vascular cerebral (AVC) são descritos problemas como rebaixamento intelectual, dificuldades na capacidade lingüística, visomotora, na organização e integração espacial. Neste estudo fizeram parte 28 crianças, sendo 14 pertencentes ao grupo experimental (GE) com diagnóstico clínico e de imagem de AVC isquêmico. Outras 14 crianças sem história de AVC que formaram o grupo controle (GC). O protocolo de investigação neuropsicológica constou de teste de inteligência, teste de coordenação viso-motor, desenho da figura humana, bateria de funções corticais e prontuário médico das crianças do GE. A análise dos resultados deste estudo mostrou que os melhores desempenhos após o AVC, estão relacionados ao menor tempo de recuperação funcional; houve desempenho superior do GC sobre o GE em todos os instrumentos utilizados, nas áreas cognitiva, perceptual e motora. Constatou-se ainda que o AVC pode levar a rebaixamento intelectual, quando ocorre um quadro de repetição do insulto vascular.

PALAVRAS-CHAVE: doença cerebrovascular, infância, avaliação neuropsicológica.

Literature shows that studies in the area of neuropsychological research with children who had cerebrovascular accident (CVA) are scarce; however, over the last years there has been a significant increase in studies which take into account neuroradiological and neurosurgical aspects¹.

In the few studies on children development after CVA, neuropsychological deficits are described²⁻⁶. Armstrong et al.⁷ assessed 194 children with sickle cell anemia, amongst whom there were 9 with past history of CVA. These children presented difficulties in linguistic and visual-motor skills, as well as in spatial organization and integration. De Schryver et al.⁵ found impaired capacity for solving problems in post

CVA children, when compared to the general population. Rodrigues et al.⁸ described children with CVA assessed evolutionally by Piaget's clinical method had present a performance significantly inferior when compared to the control group. The sequelae of a CVA during childhood are not yet as widely known as of one during adulthood; however, the more studies there are in the area, the more details shall be discovered, favoring professionals involved in the rehabilitation of such children⁹.

Based on the neuropsychological theory and specific assessment instruments, this study aimed at drawing up a neuropsychological profile of a group of children with past history of CVA.

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METHOD

Twenty-eight children participated in this study; 14 of them belonged to the experimental group (EG) with both clinical and imaging diagnostic of ischemic CVA. Children with vascular problems related to brain-skull trauma and associated to genetic syndromes were excluded. The other 14 children were placed in the control group (CG), paired according to gender and age to the EG, and besides not having a past history of CVA or another neurological problem, they presented appropriate academic performances. Two public schools of the region of Campinas (Brazil) were chosen by simple random draw, the children were selected by the pedagogue and the teacher, whose criterion was a good performance according to the grade.

The participation of each child in both groups was authorized by parents who agreed with the research by signing a Free Consent Agreement Form approved by the Ethical Committee on research of the Faculdade de Ciências Médicas (FCM) of the Universidade Estadual de Campinas (UNICAMP).

Based on the protocol for neuropsychological research on cerebrovascular disease, the following instruments were used: the medical records of each child from the EG belonging to the Clinical Hospital (CH) of UNICAMP; the Wechsler Intelligence Scale for Children - WISC¹⁰; the Visual-Motor Gestalt Test¹¹, the Luria-Nebraska Battery (LNB)¹² and the Test of human figure drawing.

RESULTS

In order to form the EG a total of 35 children were assessed – who according to inclusion and exclusion criteria were or were not selected for the study. The EG consisted of 14 children, being 7 males and 7 fe-

males; the age ranged from 7 years and 1 month to 14 years and 1 month, with an average of 9 years and 10 months. As for schooling, it ranged from 1st to 6th grades, besides special education and no schooling.

As the CG was paired according to age and gender to the EG, it was also formed by 14 children, 7 males and 7 females, and with age range during the assessment period of 7 years and 5 months to 14 years and 2 months, with average age of 9 years and 11 months. Schooling ranged from 2nd to 8th grades.

The results of the study with the total 28 children were organized in a way that relations among the instruments used as procedure of this work and also between both groups (EG and CG) could be demonstrated.

The data from the EG children (Table 1) refer to the information obtained from the medical records at the Clinical Hospital - UNICAMP, showing that the first CVA occurred between the ages of 13 months and 10 years and 6 months, being 2 children during the interval from 0 to 3 years of age; 8 children between 3 and 6 years of age; and 4 in the period from 9 to 10 years of age. There has been recurrence in 4 children. All the children presented ischemic type CVA and 2 of them had a CVA with hemorrhagic transformation. The admission to UNICAMP's Clinical Hospital happened from January of 1991 to November of 2000. Children's age during the assessment ranged from 7 years and 1 month old to 14 years and 1 month old, with average of 9 years and 10 months.

Table 1. Cerebrovascular disease: identification.

S	Gender	Recurrence	Type of CVA	CH ad.	Age during assessment	Time lag after CVA	
1	F	1y 1m	No	I	Dec-91	7y 1m	6y
2	M	6y 11m	No	I	Jan-98	9y 3m	2y 4m
3	M	4y 6m	No	I	Oct-94	10y 1m	5y 5m
4	M	3y 1m	No	I	Jan-91	10y 10m	7y 9m
5	M	5y 6m	No	I	Jun-92	11y 10m	6y 4m
6	F	9y 11m	No	I	Nov-97	12y	3 y
7	F	7y 6m	No	I → H	Mar-92	14y 1m	6y 7m
8	F	10y 6m	No	I	Feb-98	12y 4m	1y 10m
9	F	7y 8m	Yes	I	Aug-00	8y 4m	8m
10	M	4y 7m	Yes	I → H	Dez-97	7y 10m	3y3m
11	M	7 y	No	I	Nov-00	7y 8m	8m
12	F	5y 4m	Yes	I	Mar-97	8y 5m	3y
13	F	7 y	Yes	I	Jun-97	10y 4m	3y 4m
14	M	4y 10m	No	I	Jan-99	8y 1m	3y 3m

S, subject; F, female; M, male; y, year; m, month; I, ischemic; H, hemorrhagic; CH, clinical hospital; ad, admission.

Table 2. Results of the imaging and neurological exams - experimental group.

S	Hemisphere	Localization	Neurological exam	Neurological evolution
1	L	Subcortical	-	-
2	R	Subcortical and brain stem	X	Hemicorrhea L
3	R	Cortical and brain stem	X	Hemiparesis L
4	R	Subcortical	X	Hemiparesis L
5	L	Subcortical	X	Hemiparesis R
6	L	Subcortical	X	Hemiparesis R
7	R	Cortical-subcortical	X	Hemiparesis L
8	L	Subcortical	-	-
9	B	Cortical-subcortical	X	Hemiparesis R
10	L	Brain stem	X	Hemiparesis R
11	R	Cortical-subcortical	-	-
12	B	Cortical-subcortical	X	Hemiparesis L
13	L	Cortical	X	Hemiparesis R
14	L	Cortical-subcortical	X	Hemiparesis R

R, right; L, left; B, bilateral; (x) presence of symptoms; (-) absence of symptoms.

Table 3. Results of the WISC - experimental group and control group.

EG						CG					
S	Age	VIQ	EIQ	TIQ	Classification	S	Age	VIQ	EIQ	TIQ	Classification
1	7y 1m	118	125	123	Superior	1	7y 5m	124	127	128	Superior
2	9y 3m	103	97	100	Average	2	9y 3m	109	97	104	Average
3	10y 1m	97	82	87	Low average	3	10y 3m	95	89	91	Average
4	10y 10m	104	85	94	Average	4	10y 10m	94	125	109	Average
5	11y 10m	95	100	97	Average	5	12y	100	93	96	Average
6	12y	87	76	80	Low average	6	12y 1m	90	99	93	Average
7	14y 1m	79	78	76	Borderline	7	14y 2m	103	94	99	Average
8	12y 4m	91	100	95	Average	8	12y 2m	100	97	99	Average
9	8y 4m	108	106	107	Average	9	8y	131	128	133	Very superior
10	7y 10m	85	68	75	Borderline	10	7y 4m	113	104	109	Average
11	7y 8m	115	111	115	High average	11	7y 8m	115	101	95	Average
12	8y 5m	77	48	60	Mental impairment	12	8y 8m	105	108	107	Average
13	10y 4m	70	71	67	Mental impairment	13	10y 8m	109	103	107	Average
14	8y 1m	91	93	91	Average	14	8y 5m	110	118	115	High average

y, years; m, months; VIQ, verbal intelligence quotient; EIQ, execution intelligence quotient; TIQ, total intelligence quotient.

The time lag between ages when vascular insult and assessment occurred ranged from 8 months to 7 years and 9 months, revealing an approximate average of 4 years and 4 months, being 9 of the children aged under the average in the period of assessment and 5 above.

At the time of the vascular insult occurrence, the clinical symptoms presented were: 14 (100%) motor symptoms; 10 (71%) changes in speech; 8 (57%) headache; 5 (36%) vomiting; 4 (48%) generalized convulsive seizure; 3 (21%) fever; 3 (21%) consciousness changes; 3 (21%) somnolence; 4 (48%) visual diffi-

culty (impairment); 2 (14%) sialorrhea; 2 (14%) abdominal pain; and 1 (7%) had other symptoms such as dizziness, diarrhea and hypertension.

Table 2 presents information from the imaging exam and neurological evolution after vascular insult. Brain injury in the left hemisphere was found in 7 (50%) of the cases; in the right hemisphere in 5 (36%) and in both hemispheres in 2 (14%) of the cases. In 6 of these cases, the subcortical area was affected, being 2 cortical, 5 cortical-subcortical and 3 in the brain stem. The neurological examination presented alterations in 11 subjects and was normal in the

other 3 children, and it was also found right hemiparesis in 6 children, left hemiparesis in 4 subjects and left hemicorrea in only one child.

As for the cognitive assessment (WISC) of the EG, the following classification was obtained for intellectual quotient (IQ): one child at the superior intelligence range; one child at the high average intelligence range, 6 at the average intelligence range, 2 children at the low average range, 2 children at the borderline deficiency range and 2 children with results which were compatible to mental impairment.

The following is the CG classification for IQ: 1 child at the very superior intelligence range, 1 child at the superior range, 1 child at the high average range and 11 children at the average intelligence range. These results are organized in Table 3.

Figure 1 shows a comparison of the WISC results from both groups, which reveals an inferiority of the responses of EG over CG. This datum was expected since in the first group there are children with intellectual reduction.

The results of the perceptive motor development of the EG in the Gestaltic visual-motor test (Bender) were obtained under two correction systems, known as Santucci and Koppitz. Based on the results, the data were grouped together according to the correction system as follows.

The scores obtained through the Santucci system¹³ were: 8 (57.1%) children presented average perceptive motor development, 5 (37.7%) had inferior performances and 1 (7.1%) child had a score above the average for his chronological age. While in the Koppitz system¹⁴ – 11 (78.5%) children reached the average and 3 (21.4%) presented perceptive motor maturity below average. There was predominance of difficulty in the shape inversion item, followed by rotation, integration and perseverance.

The CG presented the following results, which also refer to gross scores from the two correction systems of the Bender test. The perceptive-motor development of this group shows the following pattern: in the Santucci system – 8 (57.1%) children with average development; 4 (28.5%) above the average and 2 (14.2%) with perceptive development below the

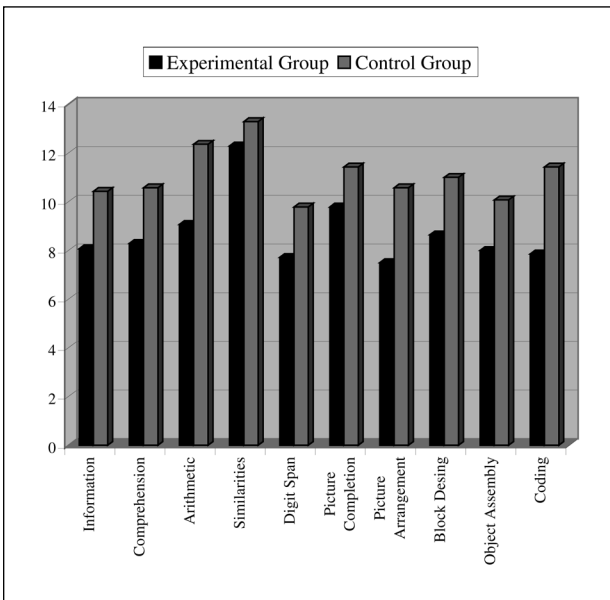


Fig 1. Comparison of the WISC results from - experimental group and control group.

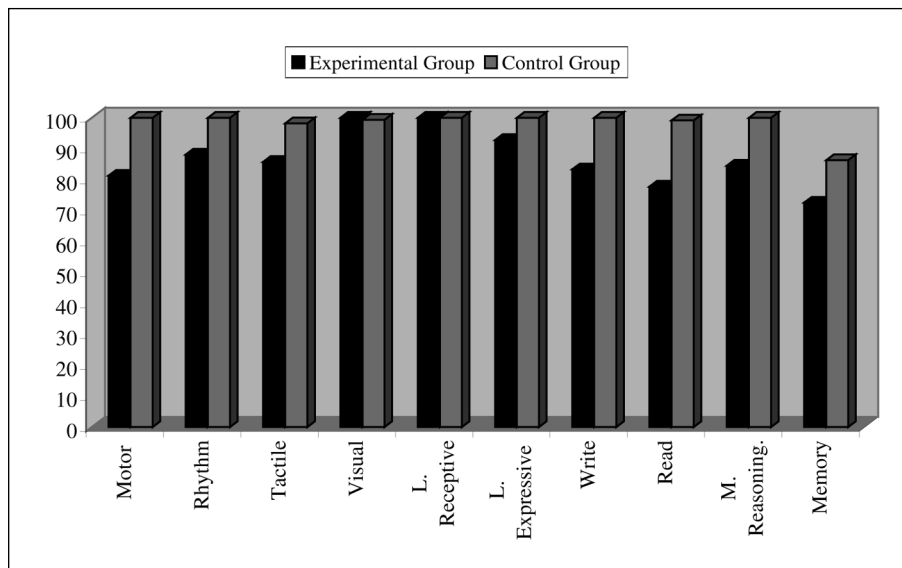


Fig 2. Comparison of the results of the LNB - experimental group and control group.

expected for the age. And in the Koppitz, 14 (100%) of the children presented average pattern for perceptive-motor development. Rotation was the most difficult item, followed by shape inversion, integration and perseverance.

In both groups, there has been a concurrence of 85.7% in the correction systems used.

The correct marks obtained in each function of the LNB from EG and CG were transformed into percentage of correct marks, as it may be noticed in figure 2, comparing both groups. As for the statistical analysis, significative results from CG over EG were found in the following functions: motor skill, tactile skill, writing, reading and memory.

The following results refer to conceptual maturity of the human figure drawing for both EG and CG population. For the EG the percentile variation ranged from 1 to 100 while in the CG it ranged from 22 to 100, increasing in this way the classification of the CG over the EG.

The following classificatory distribution was found in the EG: 2 (14.2%) much superior performances; 1 (7.1%) superior score; 3 (21.4%) above average; 4 (28.5%) average performances, 2 (1.2%) borderline performances and 2 (1.2%) with impaired performances.

While in the CG the following data were found: 3 (21.4%) much superior scores; 5 (35.7%) superior scores; 1 (7.1%) above the average performance; 4 (28.5%) average performances; and 1 (7.1%) borderline performance regarding the maturity of the human figure drawing.

DISCUSSION

CG's superior performance in the LNB was significant in the following functions: motor and tactile skills, writing, reading and memory. The motor sequelae in the EG altered the capacity for both global and fine motor coordination, what hindered successful performances in such activities. There was no direct relation between the worst performances and cognitive capacity but with motor difficulty, more specifically with the hemiparesis condition.

As for the tactile skill both groups presented significant differences. The left-right domain, mediated in this test, showed the maturity function to be delayed, and according to Le Bouch¹⁵ the stabilization of the lateral function occurs between 6 and 8 years of age. The statistical significance of CG over EG for writing and reading skills can be justified by factors such as: difficulty in management of the learning-teaching process by both parents and teachers;

longer schooling time of the CG and intellectual reduction of the EG.

There was difference between EG and CG in relation to memory ability, mainly in the exercise of immediate auditory memory (word), which made it possible to correlate the results of this test to the digit span subtest, and EG presented low scores in both tests. Regarding neuropsychological mechanisms of memory organization, Barbizet and Duizabo¹⁶ reveal that the verbal information quantity (verbal or visual span) which the individual is able to memorize after a single exposition is limited. This capacity demands the integrity of functional areas; immediate memory is decreased constantly in cases of damaged cortical areas.

In the works of Lefèvre¹⁷ and Tabaquim¹⁸ with children, the first related to Moyamoya disease and the second about hemiparetic cerebral palsy and learning disorders, the similarity subtest was the one that presented the best performances. The same thing may be observed in both groups of this study; this subtest demands conceptual thought capacity and it is less influenced by social or formal education than other verbal subtests¹⁹.

The intellectual deficit found in the EG of this study is superior when compared to the general population; however, these indexes may not be considered permanent, once the capacity of cerebral reorganization was clearly observed, and adding to this conclusion, there is the fact that remarkable changes in IQ are common in pediatric age groups²⁰, as well as in all age groups.

Vargha-Khadem²¹ reported that cognitive reduction is more common when the CVA occurs between the ages of 2 and 4 years old, but at this present study this finding was not confirmed. Cognitive reduction occurred in children who had CVA between the ages of 5 and 7 years old, which can have been influenced by repetition of the vascular condition.

By analyzing the subjects with intellectual reduction, some variations as for the performances with in each instrument may be noticed. However, there were constant low scores in the tests, suggesting that the CVA recurrence can be quite destructive to cerebral tissues, which makes it difficult for reorganization to occur. Still on the recurrence of vascular insult, for subject S9, at the time of the insults, the abstraction functions (mathematics, reading and writing) were already grouped together and there were no significant losses after the CVA, differently from subjects S10, S12 and S13 (with injury in the left hemi-

sphere) who had not acquired yet or were still in process of acquisition of these functions.

According to Wood et al. in Oliveira²² between the ages of 5 and 8 years old, the left hemisphere becomes more specialized for language, regarding the development of reading and writing functions. Pitchford also cited by Oliveira²² presented a five children study with CVD in the left hemisphere, being that two of them were in pre-school age. Damage in verbal performance with signs of reading and writing difficulty was observed. For Luria²³ the damages are greater when there are lesions in the primary area, yet lesions of secondary areas imply participation of other functional circuits for functional recovery.

Bender's constructive visual task, through perception and responses to stimuli, does not constitute a simple process, but involves neuropsychological aspects of cerebral functioning¹⁵. The performance (EG) in the gestaltic visual-motor test, in both correction systems, revealed that reception, integration and expression of information were being processed appropriately.

Antunha²⁴ explains that the development of internal image of one's own body results from the somesthetic projection (somesthetic and parietal-occipital cortices). In this test, human figure drawing, a good performance was observed in the EG, but it was noticed that the CG has a better group performance, suggesting that both groups present an appropriate representation of body image or scheme.

One aspect that deserves attention is the cognitive performance related to gender. Studies have shown that gender differences change cerebral processing, that is, men and women have different cognitive styles^{25,26}.

It may be observed that in this study it was possible to form EG with seven girls and seven boys. Through their performances as a subgroup, the females are found with two individuals presenting cognitive performances that are compatible to borderline deficiency and mental impairment. While the males present only one borderline deficiency individual. Another important point is the fact that the female subgroup presents more individuals with CVA recurrence in relation to the male one, a 3:1 ratio.

A valuable analysis of the relation between gender and abilities in the population studied here is considered to be impracticable due to cognitive reduction observed in the female subgroup of the EG. Such finding affects the performances of this subgroup incisively. Nevertheless, it may also be noticed

that in this study the females with a past history of vascular insult present greater possibilities of intellectual reduction and CVA recurrence.

In this study, 4 years and 4 months was the average time span between vascular insult and assessment. It was not possible to establish stronger strictness for the control of this variable due to the low number of casuistic. Out of five children who presented above the average performances only one showed borderline cognitive performance, while the others had normal variation of intelligence. As for the other nine children evaluated who were under this average, two presented evolution with intellectual performance compatible to mental impairment, borderline deficiency and the others with normal variation of the cognitive capacity. Moreover, four children had a recurrence of the vascular insult.

In summary, it may be suggested that the children who were assessed above the average of 4 years and 4 months after the CVA had better cerebral reorganization in response to the lesion, revealing more adaptative cognitive and behavioral resources. However, it must be observed that for the children who were assessed under this average the recurrence factor denotes the severity of cerebral damage, once the repercussion of the lesion impact seems to diminish possibilities of plasticity, which may be noticed from behavioral expressions that are or are not in accordance with the age group to which the child belongs. Mello and Muszkat²⁷ reveal that a developing brain is less vulnerable to the effect of a lesion. The present study shows the importance of specific evaluations in groups of post-CVA children, because neuropsychological deficits which hinder the individual's satisfactory development were found. The parameters of a child development after a CVA are still being grounded, yet it is believed that some contributions in order to reach them could be substantiated in this work.

REFERENCES

1. Bowen M, Marks MP, Steinberg GK. Case report: neuropsychological recovery from childhood moyamoya disease. *Brain Dev* 1998;20:119-123.
2. Koelfen W, Freund M, König S, Varnholt V, Rohr H, Schultze CH. Results of parenchymal and angiographic magnetic resonance imaging and neuropsychological testing of children after stroke as neonates. *Eur J Pediatr* 1993;152:1030-1035.
3. Ganesan V, Isaac E, Kirham FJ. Variable presentation of cerebrovascular disease in monovular twins. *Dev Med Child Neurol* 1997;39:628-631.
4. Ciasca SM, Alves HL, Guimarães IE, et al. Comparação das avaliações neuropsicológicas em menina com doença cerebrovascular bilateral (Moyamoya) antes e após a intervenção cirúrgica. *Arq Neuropsiquiatr* 1999;57:1036-1040.
5. De Schryver ELM, Kappelle LJ, Jennekns-Schinkel A, et al. Prognosis of ischemic stroke in childhood a long-term follow-up study. *Dev Med Child Neurol* 2000;42:313-318.

6. Guimaraães IE, Ciasca SM, Moura-Ribeiro MLL. Neuropsychological evaluation of children after ischemic cerebrovascular disease. *Arq Neuropsiquiatr* 2002;60:386-389.
7. Armstrong FD, Thompson RJ, Wang W, et al. Cognitive function and brain magnetic resonance imaging in children with sickle cell disease. *Pediatrics* 1996;97:864-870.
8. Rodrigues SM, Ciasca SM, Moura-Ribeiro MVL. Ischemic cerebrovascular disease in childhood: cognitive assessment of 15 patients. *Arq Neuropsiquiatr* 2004;62:802-807.
9. Ganesan V, Hogan A, Shack N, Gordon A, Isaacs E, Kirkham FJ. Outcome after stroke in childhood. *Dev Med Child Neurol* 2000;42:455-456.
10. Wechsler D. Manual for the Wechsler intelligence scale for children. New York: Psychological Corporation, 1974.
11. Bender L. Teste gestáltico visomotor. Buenos Aires: Paidós, 1964.
12. Golden CY. The nebraska neuropsychological children's battery. In: Reynolds CR, Fletcher-Jensen E (eds). Handbook of clinical child neuropsychology. New York: Plenum Press, 1989:193-204.
13. Zazzo R. Manual para exame psicológico da criança. São Paulo: Editora Mestre Jou, 1968.
14. Cunha JA. Psiodiagnóstico-R. 4ed. Porto Alegre: Artes Médicas, 1993.
15. Le Bouch J. O desenvolvimento psicomotor: do nascimento até 6 anos. 6.Ed. Porto Alegre: Artes Médicas, 1992.
16. Barbizer J, Duizabo Ph. Manual de neuropsicologia. Porto Alegre: Artes Médicas; São Paulo: Masson, 1985.
17. Lefèvre BH. Neuropsicologia infantil. São Paulo: Savier, 1989.
18. Tabaquim MLM. Avaliação neuropsicológica: estudo comparativo de crianças com paralisia cerebral hemiparética e distúrbios de aprendizagem. Tese, Campinas, 2002.
19. Lezak MD. Neuropsychological assessment. New York: Oxford Universit Press, 1983.
20. Levin HS, Soukup VM, Benton AL, Fletcher JM, Satz P. Avaliação neuropsicológica e intelectual de adultos. In: Kaplan H, Sadock BJ (eds). Tratado de psiquiatria. 6.Ed. Porto Alegre: Artmed, 1999:613-633.
21. Varga-Khadern F. Neuropsychological outcome (Abstr). In 8th International Child Neurology Congress. Slovenia, 1998:282-283.
22. Oliveira KT. Distúrbio adquirido em crianças com doença cerebrovascular: aquisição de linguagem em lactentes e pré-escolares. Dissertação. Campinas, 2002.
23. Luria AR. Fundamentos de neuropsicologia. São Paulo:Edusp, 1981.
24. Antunha ELG. Avaliação neuropsicológica na infância. In Oliveira VB, Bossa ANA. (org). Avaliação psicopedagógica da criança de zero a seis anos. 2.Ed. Petrópolis: Editora Vozes, 1994:89-122.
25. Kolb B, Whishaw IQ. Neurociência do comportamento. Barueri:Manole,2002.
26. Goldberg E. O cérebro executivo: lobos frontais e a mente civilizada. Rio de Janeiro: Imago, 2002.
27. Mello CB, Muszkat. Reabilitação cognitiva e a lesão cerebral na infância: uma perspectiva multidisciplinar. *Temas Sobre Desenvolvimento* 2004;13:5-12.