

MOTOR PERCEPTUAL FUNCTIONS OF CHILDREN WITH CLEFT LIP AND PALATE

Funções percepto-motoras de crianças com fissura labiopalatina

Funciones perceptivo-motrices de niños con fisura labiopalatina

Original Article

RESUMO

Objetivo: Investigar as funções neuropsicológicas perceptuais visomotoras de crianças com fissura labiopalatina. **Métodos:** Estudo observacional, prospectivo, transversal, descritivo, realizado em um hospital de referência para anomalias craniofaciais no período de agosto/2012 a fevereiro/2013. Participaram 50 crianças de ambos os sexos, 25 com fissura labiopalatina compondo o GI e 25 como grupo controle, GII, com idade de 10 anos. Utilizaram-se para a avaliação os testes Matrizes Progressivas Coloridas, Gestáltico Visomotor e as subprovas de Exame Neuropsicológico, seguidos de aplicação estatística, utilizando-se testes de promoção e correlação (Fischer) ao nível de significância de 5%. **Resultados:** O grupo com fissura labiopalatina demonstrou desempenhos abaixo da média esperada para a idade, sendo o grupo com o tipo transforame labial o mais prejudicado na performance da competência perceptual visomotora. Os tipos de fissura pré e pós-forame obtiveram escores na média, com pontuações pareadas ao grupo controle. **Conclusão:** As crianças com fissura labiopalatina do tipo transforame mostraram-se mais vulneráveis a alterações nas funções visomotoras. Além disso, as habilidades nos domínios perceptivos construtivos estão relacionadas a desempenhos classificados abaixo da média esperada para a faixa etária na população-alvo.

Descritores: Neuropsicologia; Fissura Palatina; Percepção.

ABSTRACT

Objective: To investigate the neuropsychological visual-motor perceptual functions of children with cleft lip and palate. **Methods:** Observational, prospective, cross-sectional, descriptive study, held at a referral hospital for craniofacial anomalies in the period from August 2012 to February 2013. The study included 50 children of both sexes, 25 of them with cleft lip and palate, comprising GI, and 25 children as control group, GII, aged 10 years. For evaluation, the study applied the Coloured Progressive Matrices, the Visual-Motor Gestalt Test, and Neuropsychological Subtests Examination, followed by statistical evaluation, using promotion and correlation tests (Fischer) at 5% significance level. **Results:** The group with cleft lip and palate showed performance below the expected average for their age, the group with the trans-incisive foramen cleft type being the most affected in the performance of visual-motor perceptual competence. Pre- and post-foramen cleft types obtained average scores, with values matching the control group. **Conclusion:** Children with cleft lip and palate of trans-foramen type were found more liable to present disorders in visual-motor functions. Furthermore, skills in the perceptual and constructive domains are related to performances classified below the expected mean for the age range in the target population.

Descriptors: Neuropsychology; Cleft Palate; Perception.

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RESUMEN

Objetivo: Investigar las funciones neuropsicológicas perceptivo-motrices de niños con fisura labiopalatina. **Métodos:** Estudio observacional, prospectivo, transversal, y descriptivo realizado en un hospital de referencia en anomalías craneofaciales entre agosto/2012 y febrero/2013. Participaron 50 niños de ambos sexos, 25 con fisura labiopalatina en el GI y 25 en el grupo control, GII, y edad de 10 años. Se utilizó los testes de Matrices Progresivas de Colores, Gestáltico Visomotor y las subpruebas de Examen Neuropsicológico en la evaluación seguidos de la estadística utilizándose las pruebas de promoción y correlación (Fischer) con un nivel de significación del 5%. **Resultados:** El grupo con fisura labiopalatina demostró desempeños abajo de la media esperada para la edad siendo el grupo con el tipo transforamen labial el más perjudicado en la actuación de la competencia perceptual visomotora. Los tipos de fisura pre y pos-foramen tuvieron puntuaciones dentro de la media con las puntuaciones pareadas con el grupo control. **Conclusión:** Los niños con fisura labiopalatina del tipo transforamen fueron más vulnerables de alteraciones de las funciones visomotoras. Además, las habilidades de los dominios perceptivos constructivos están relacionadas con desempeños clasificados abajo de la media esperada para la franja de edad de la población investigada.

Descriptor: Neuropsicología; Fisura del Paladar; Percepción.

INTRODUCTION

Perception can be defined by the entry of sensory impression in the consciousness, helping the subject to build an image of themselves and the environment that surrounds them. This process of transforming reality results in subjective, personal impression, involving the mobilization and operation of the central brain mechanisms^(1,2).

For the interpretation of sensory signals to take place in the processing of visual information, it demands the activation of the mechanisms of attention, so that the subject becomes aware of the surrounding stimuli, recognizing and/or comparing them to the information already stored in memory. That way, the individual can process information, make decisions, and act according to what they feel most appropriate to the situation⁽³⁾. Visual perception is a cognitive function through which the information on the visual environment becomes available in the consciousness⁽⁴⁾, being one of the main routes for transmission and access to information that is later used in the construction of mental representations and one of the most important senses in children's neurological and psychomotor development⁽⁵⁾.

Recent investigations in the field of Psychology, with neuropsychological studies of children with cleft lip and palate, have shown impairments in several cortical

functions, including those related to attention, perception, language, and memory, with consistent impact on school learning⁽⁶⁻¹⁰⁾.

Although traditional literature tends to give an environmental explanation to behavior, learning, and neuropsychological features⁽¹¹⁾, more recent studies have demonstrated, through brain imaging, greater biological interaction, based on neuronal and direct differences in neuromaturation in children with cleft lip and palate⁽¹²⁻¹⁴⁾.

The central nervous system (CNS), fundamental to the development of the craniofacial complex, emerges from the neural plate, which curves inward to form the neural tube, as its enlarged anterior end is segmented into three vesicles, thus leading to the development of forebrain (prosencephalon), midbrain (mesencephalon), and hindbrain (rhombencephalon). The expansion of the forebrain gives rise to the frontonasal process, and the neural crest cells, derived from the middle and posterior brain, migrate to the area of the pharyngeal arches, which are closely related to the development of the middle and lower regions of the craniofacial complex^(15,16). Midline defects, mostly caused by incomplete differentiation, include CNS anomalies (corpus callosum and cerebellar abnormalities etc.), median facial clefts, clefts of lip/palate/jaw/larynx, among others⁽¹⁷⁾, and are liable to lead to several facial phenotypes.

One of the most well-documented findings in literature on longitudinal outcomes of children with cleft lip and palate (CLP) is the increased risk for language disorders (affecting receptive, expressive, semantic, pragmatic, or verbal memory abilities), with impairment severity associated with the length of the cleft palate⁽¹⁸⁻²⁴⁾. Recent research findings provide prospective evidence of neurobiological association between cleft type and neuropsychological, academic factors, and distinct behavioral profiles^(13,25).

Recent studies in the field of Psychology, with neuropsychological investigation of children with cleft lip and palate at school age^(6-9,12,13), have shown losses in several cortical functions, including those related to attention, perception, language, and memory, with consistent impact on school learning. The child in elementary school experiences requirements for the learning process and demands the development of more effective cognitive abilities.

Literature provides few studies on the neuropsychological cognitive functions of children with this anomaly. Therefore, studies that seek this apprehension may represent an important contribution towards understanding the learning of children with cleft lip and palate and thus help to develop prevention and rehabilitation programs, enabling greater educational and social insertion, and minimizing conflicts possibly arising from prejudice generated by the physical appearance.

Comparing the verbal and nonverbal performances of children with different types of CLP can contribute to greater understanding of the condition and provide specific rehabilitation parameters for this population. With this view, this study aimed to investigate the neuropsychological, visual-motor perceptual functions of children with cleft lip and palate.

METHODS

This is a cross-sectional, prospective, observational and descriptive study, with 50 participants of both sexes, aged from 10 years to 10 years and 11 months, held at the Craniofacial Anomalies Rehabilitation Hospital of the University of São Paulo (*Hospital de Reabilitação de Anomalias Craniofaciais da Universidade de São Paulo - HRAC -USP*) and at Mercedes Paz Bueno State School, both in the city of Bauru, São Paulo, Brazil, from August 2012 to February 2013.

Two groups were formed: GI, comprising 25 participants with cleft lip and palate, 5 of them with pre-foramen cleft type, 5 post-foramen, and 15 trans-foramen; and GII, with 25 participants from public schools, without other changes, composing the control group.

The inclusion criteria for GI were: being within the age range proposed for the study, from 10 years to 10 years and 11 months; being enrolled in HRAC-USP; presenting pre-foramen, post-foramen, or trans-foramen cleft lip and palate, whether unilateral or bilateral; intellectual level on average (III), below (IV), or above (I and II) average; without neurological diagnosis, intellectual impairment, or sensory deficits. Inclusion criteria for GII: being within the age range proposed for the study, from 10 years to 10 years and 11 months; being enrolled in regular public elementary school; intellectual level on average (III), below (IV), or above (I and II) average; without neurological diagnosis, intellectual impairment, or sensory deficits.

For evaluation, the following tests were used: Raven's Coloured Progressive Matrices⁽²⁶⁾, which evaluates the overall intellectual level, by means of three series, each with 12 pictographic problems, in which the child must deduce the relationships and extract the meaning of a conflicting situation; Bender Visual-Motor Gestalt Test⁽²⁷⁾, which aims to assess the child's visual-motor maturity through the identification of distortions in the perceptual-motor graphic layout, regarding the angle, orientation, and relative position; Neuropsychological examination^(6-8, 28), consisting of ten exams, with specific subtests, aimed at the analysis of higher cortical functions in 7- to 12-year-old children. In the present study, the visual perception tests were selected, including four subtests: 1) Perception of

objects and drawings; 2) spatial orientation; 3) intellectual operations in space; and 4) visual retention and recovery.

The scoring and interpretation criteria were adopted according to the regulations of each instrument. Angles, spatial orientation, and relative position were considered in the visual-motor subtests.

After the accomplishment of research ethical procedures involving the participants and their parents or guardians (signature of Informed Consent Form), the tests were applied by an evaluator, psychologist and author of this study, with expertise in instrumentation of the material. The tests were performed in previously arranged rooms, in the facilities of the participating institutions, under proper control of variables (light, noise, and privacy) that were likely to interfere in the proceedings. After their application to the participants, the obtained data was recorded in specific protocols, without the presence of parents/guardians, and organized into sheets.

Described in absolute (n) and relative (%) numbers, data was subjected to statistical analysis, using ratio and correlation tests (Fisher), adopting significance level at 5% to assess statistically significant differences.

This research had its project approved by the Ethics Committee for Research on Human Beings of the Craniofacial Anomalies Rehabilitation Hospital (HRAC-USP), Opinions No. 86443/2012 and No. 262,625/2013, in accordance with resolution No. 466/2012 of the National Health Council, and the participating children had their Consent Forms signed by their parents or guardians.

RESULTS

Regarding the characterization of GI, there was a predominance of females (n=14; 56%), enrolled in the 5th year of elementary school (n=13; 52%). In GII, male participants were the majority (n=18; 72%), also mostly attending the 5th year of elementary school. As for the type of cleft, there was a predominance of participants with trans-foramen cleft (n=15; 60%).

As for the evaluation of the intellectual level, 19 (76%) participants in GI had scores on average, and 6 (24%) were above the average; in GII, 10 (40%) were on average, and 15 (60%) were above. According to eligibility criteria for the current study, no participants had scores below the average; the intellectual level was thus stratified by convenience.

In the visual perception tests of the Neuropsychological Examination, out of 25 participants in GI, 10 (40%) answered correctly 100% of the questions, being above the expected average for this age; 11 (44%) were on average, and 4 (16%) scored below the mean (considered below with 20% of accuracy). The results of both groups demonstrated

ability in the perceptual function, given the performances found on average and above the average: in tasks related to “drawing design” (n=21; 84% of GI participants), “objects in the drawing” (n=20; 80% of GI participants), and “spatial orientation” (n=18; 72% of GI participants); in GII, 23 (92%) of the participants reached that performance, in all three tasks.

The performance on perceptual cognitive activities, classified on average in the “spatial operations”, was lower for both groups: 13 (52%) in GI and 20 (80%) in GII. Table I shows the number of participants in the tests of visual perception with scores on average, above and below the average expected for their age.

On the characterization of the visual perception through Neuropsychological Examination, according to cleft types, it was observed that 23 (92%) subjects reached scores above the expected average (Table II). The subtest “intellectual operations in space” was found to be the most troublesome for participants with pre-foramen cleft, compared to other types.

When the visual-motor subtest was analyzed in GI, in relation to angles construction, orientation and relative position, the overall result indicated 6 (24%) participants with score above the average, 7 (28%) on average, and 12 (48%) below. In the subtest involving angles, 10 (40%) subjects performed above average, 7 (28%) on average, and 8 (32%) below. In spatial orientation, only 2 (8%) subjects were above average, 9 (36%) on the average, and 14 (56%) below. In the subtest on relative position, 10 participants obtained score above the expected average, 8 were on average, and 7 were below the average (Table III).

In GII, the subtest on angles resulted in 7 participants above average, 13 on average, and 5 below. In the spatial orientation subtest, 3 were above average, 12 on average, and 10 below. In the subtest on relative position, 6 participants scored above the expected average, 4 on average, and 5 below average (Table III). Thus, comparing the two groups descriptively, there was a discrepancy regarding visual-motor maturity. GI had fewer subjects (n=13; 52%) on the average scoring and above it, evidencing in the sample

Table I - Number of participants in classifications attained in visual perception tests of the Neuropsychological Examination in GI (group I) and GII (group II). Bauru, SP, 2012-2013.

Classification	Figure drawing		Objects in drawing		Spatial orientation		Operatios in space	
	GI	GII	GI	GII	GI	GII	GI	GII
Above average	10	18	08	15	06	13	03	12
On average	11	05	12	08	12	10	10	08
Below average	04	02	05	02	07	02	12	05

GI: Group I; GII: Group II.

Table II - Visual perceptual results of GI (group I) according to the types of clefts. Bauru, SP, 2012-2013.

Cleft Type	Figure drawing (%)			Objects in drawing (%)			Spatial orientation (%)			Operations in space (%)		
	Pre	Post	Trans	Pre	Post	Trans	Pre	Post	Trans	Pre	Post	Trans
	Above average	80	100	93	100	80	73	60	60	80	40	60
On average	0	0	0	0	0	0	0	0	0	0	0	26
Below average	20	0	7	0	20	27	40	40	20	60	40	27

Pre: pre-foramen; Post: post-foramen; Trans: trans-foramen.

Table III - Results of visual-motor subtests in GI (group I) and GII (group II). Bauru, SP, 2012-2013.

Subtests	GI			GII		
	Below average	On average	Above average	Below average	On average	Above average
Angle	08	07	10	05	13	07
Orientation	14	09	02	10	12	03
Relative Position	07	08	10	05	14	06
Global	12	07	06	08	03	14

GI: Group I; GII: Group II.

a representative contingent of participants with global visual-motor immaturity (n = 12; 48%) compared to the control group (n=17; 68%). When the subtests results were analyzed, “spatial orientation” was found to be the ability with more deficits evidenced in GI (n = 14; 56%). However, the statistical analysis with use of Fisher’s exact test showed no statistically significant correlation in the comparison between groups (Table IV).

By analysing the visual-motor results according to the different types of cleft in GI, the results were below the average in all sub-items (global, angles, spatial orientation, and relative position) in participants with the trans-foramen type of cleft (n=16, 67 %; n=10, 40%; n=16, 67%; n=10, 40%; respectively, for the said sub-item). In the perception “angles”, better performance was found in the pre-foramen

cleft group (n= 20, 80%; n=25, 100%, n=20, 80%; n=25, 100%; respectively, in relation to that sub-items), as shown in Table V.

Comparison of GI and GII results in relation to the overall score of visual-motor tests yielded the following scores: 7 (28%) participants in GI and three (12%) in GII were on average; 3 (12%) participants in the GI and 8 (32%) in GII were below average; and 5 (20%) in GI and 14 (56%) in GII were above average.

Thus, as for the different types of cleft within GI, visual-motor and visual perceptual results showed that subjects with trans-foramen cleft had below-average results in all sub-items of the instruments used, including the overall score.

Table IV - Comparison of groups and number of subjects according to classifications (on, above, and below average) in the results of Neuropsychological Examination in GI (group I) and GII (group II). Bauru, SP, 2012-2013.

	Classification			<i>p</i>
	Above average	On average	Below average	
Group I	10 (40.00%)	8 (32.00%)	7 (28.00%)	0.94
Group II	8 (32.00%)	10 (40.00%)	7 (28.00%)	

*p value

Table V - Global and partial results in visual-motor perceptual subtests in different types of cleft. Bauru, SP, 2012-2013.

Cleft Type	Global (%)			Angles (%)			Spacial Orientation (%)			Relative Position (%)		
	Pre	Post	Trans	Pre	Post	Trans	Pre	Post	Trans	Pre	Post	Trans
Above average	40	40	13	40	60	33	0	20	6	60	80	20
On average	40	40	20	60	0	27	80	20	27	40	0	40
Below average	20	20	67	0	40	40	20	60	67	0	20	40

Pre: pre-foramen; Post: post-foramen; Trans: trans-foramen.

DISCUSSION

Children with cleft lip and palate are more vulnerable to distortions in the interpretation of sensory signals in the processing of perceptions⁽²⁹⁾. Such changes are likely to affect the essential skills of learning processes, giving rise to speech and auditory processing disorders, and difficulty in socialization, resulting from the very fissure conditions. Consequently, the association with school failure is evidenced in these children⁽³⁰⁻³²⁾.

The group with cleft lip and palate (GI), aim of this study, was composed of participants with preserved intellectual level, without syndromic conditions or other

associated developmental deficits. Although the control group had more participants ranked above average than GI, the sample did not include participants with intellectual impairment, which could represent an important bias in affected cognitive tests. The intellectual conditions of GII, however, did not represent evidence of major discrepancies in performance in the assessed cognitive tasks compared to GI.

This study evaluated the visual perception of children with cleft lip and palate (GI), compared to the control group without that condition (GII). In GI, regarding the intellectual capacity, by using spatial and temporal reasoning, 19 subjects presented results on the expected average, while

6 subjects were above the average. That competence could represent a guarantee of good performance in visual-spatial tasks, which did not happen in the current study, thus making it possible to understand the involvement of different processing routes^(2,12,23). Non-verbal spatial-temporal reasoning for the understanding of visual stimuli involves temporo-occipital cortical pathways, and building responses involves connections between motor areas, supplementary motor areas, and the prefrontal cortex⁽²⁾. Moreover, the inhibitors systems are implicated, as well as reticulum systems involved in energy metabolism, both modulating sensitivity, posture, and tone, ensuring programming and planning of the sequence of actions demanded for the accomplishment of the task^(2,33).

In this study, visual-motor performance was below average in GI, mainly due to low scores obtained in subtest "orientation", which assesses the children's skills to respond in terms of perceptual and motor spatial organization. In a study⁽³³⁾ with 190 10-year-old children, referred for clinical and psychological treatment with complaints about low academic performance, 81.1% had below-average results in visual-motor tests similar to those used in this study, but the errors were mainly found as distorting shape and rotation, which typifies difficulties related to the orientation of the figure in the test. Other study⁽³⁴⁾ held with adolescents with cleft lip and palate identified in the sample difficulties in skills related to the accomplishment of visual-motor perceptual tasks and verified the effectiveness of a remediation program in the rehabilitation of these affected skills. The results of the current study corroborate the identification of impaired visual-motor functions, with errors most frequently found in the item "orientation".

Taking into account that motor skills represent the basic process of adaptation and learning, through which the individual can assimilate and accommodate the knowledge⁽³⁵⁾, the lack of integration between neurological organization and tone regulation can be an interfering factor the way children perform both their recreational or academic activities, whether in the construction of angles, spatial orientation, or relative positioning in copies of figures⁽³⁵⁾. In this study, the preserved intellectual levels and the overall visual perception at satisfactory levels in GI allowed to infer that the preserved trophic resources, in this sample, are favourable to optimized academic performances.

Participants with the trans-foramen type of cleft lip and palate had the most impaired performances in visual-motor subtests in this study. This type of cleft is one of the most complex orofacial changes, which brings malocclusion problems and limitations in feeding, swallowing, breathing, and in speech^(36,37). Whereas the evolution and motor control, not only of the orofacial segment, but of the whole individual, depend on the maturation of the nervous system

and on sensory and motor experiences, children with limiting changes can be deprived of such maturation^(35,36).

Acquisition of motor skills is integrally linked to the development of awareness of the body, space, and time, which constitute basic domain components for both motor learning and for activities performance in school education⁽³⁸⁾. Children with pre-foramen cleft, whose deformity is restricted to the primary palate, undergo reconstructive surgeries from three months of age on, having their entire anatomical lip structure reconstituted, favouring rehabilitation, psychosocial insertion, and the consequent adjustment in academic skills development⁽³⁸⁾.

The skills to build "angles", important in formal learning of writing, were analyzed in this study by means of the visual-motor tests. The best performance was found in participants with pre-foramen cleft, compared to other types. These findings corroborate the results of other studies^(6,13,33).

Among the neuropsychological functions, visual perception is crucial to academic achievement and a major resource for the child's neurological development⁽³⁹⁾. The subtest "operations in space" of the Neuropsychological Examination, involving perception and motor skills, was the most troublesome for both groups in this study, and a great part of the participants (48%) in GI were classified below the average. A research⁽⁶⁾ developed with children with cleft lip and palate assessed the graphic-perceptual-motor skills related to visual-spatial function and identified deficits in 58% of the participants, corroborating the lower classification evidenced in this study. The activities demanded self-regulation and self-direction of the child in the execution, with planning and control of systematic movements for the accomplishment of the task. That way, the visual-motor perceptual cortical functions proved immature.

This study showed that participants with trans-foramen cleft had below-average performances in all visual-motor subtests and in those within the Neuropsychological Examination that are related to spatial orientation and operations in space. As the trans-foramen cleft has the full and simultaneous involvement of the primary and secondary palates, extending from the lip to the uvula, and across the alveolar ridge, there are greater anatomical and functional impairment⁽³⁴⁾, requiring constant support for the realization of certain activities, including handling. In result, the development of visual-motor skills for acquirement of independence in daily life appears sometimes hindered in its efficiency and, therefore, in activities requiring visual-motor integration, performance may prove to be lower than expected for the age. These findings were similar to those obtained in another study⁽⁷⁾ about visual-constructive dyspraxia in 77 children with cleft lip and palate; its

results pointed to difficulties in tests of spatial orientation, indicating deficits in neuropsychological visual-constructive perceptual skills.

It is expected that the results of this study may contribute to enhance the available knowledge on the cognitive functioning of children with cleft lip and palate, enable greater understanding of the evidenced difficulties and, in consequence, promote interventions at earlier ages in relation to the elementary school education.

Although deficits in visual-motor perceptual skills of children with CLP are evident, there are still uncertainties regarding a likely influence of the cleft type on the deficit patterns. The cognitive development of individuals with cleft lip and palate focusing on the neuropsychological functions is still little investigated in the specific literature we reviewed and should be assessed within the public health to assist thousands of children facing such condition.

In order to identify losses arising from the underlying disease and others due to cognitive difficulties and environments lacking proper stimulation, the differential diagnosis will conduct guidelines and interventions adjusted to formal learning of children with cleft lip and palate. It is important that new research take place, with a more significant number of participants, to foster interest in the subject, and thus develop studies that may provide more conclusive answers.

Deficient resources and mechanisms of sensorineural, neuromotor and/or neurolinguistic nature influences on the cognitive, affective, and social development of children, impacting on educational and social aspects of school learning. The existence of scientific evidence showing the contribution of health to the quality of life of individuals with cleft lip and palate is undeniable. It is also known that aspects of the social life are contributing factors to health promotion.

However, it takes more than access to quality medical care services, as it requires healthy public policies and an effective intersectoral coordination of government and the conscious mobilization of the general population in the adjustment of subjects with such abnormalities. The birth of a child with anomaly could destabilize both the child and the family, with unfulfilled expectations and resulting conflicts in its structure and functioning.

Thus, the health team treating children with CLP, including the Neuropsychology professionals, play an important role in child development, support for families, and promoting adherence to treatment. This involves a professional performance that takes health integrality as a reference, with the concept of humanization of care, and the user embracement by the staff, providing support from birth

until the end of the treatment process, and in subsequent monitoring as well.

This includes the potential demands of the child enrolled in the school system, with academic learning and the need for actions involving the family and school institution. Thus, the identification of inherent resources of children with CLP can be a very significant differentiator in building their self-esteem and providing them instruments for better adjustment facing the challenges of learning in the academic context.

CONCLUSION

This study showed that children with trans-foramen type of cleft were more vulnerable to changes in visual-motor functions. In addition, it was found that the skills in construction and perceptual domains are related to performance ranked below the expected average for this age group in the target population.

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