Apraxia da fala na infância em foco: perspectivas teóricas e tendências atuais****

Childhood speech apraxia in focus: theoretical perspectives and present tendencies

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****Trabalho Realizado na Faculdade de Fonoaudiologia de Alagoas da Universidade Estadual de Ciências da Saúde de Alagoas.

Artigo de Revisão de Literatura

Artigo Submetido a Avaliação por Pares

Conflito de Interesse: não

Abstract
Background: the appearance of the praxis disorder during the firsts years of development has been frequently designated as chidhood apraxia of speech. The present theoretical perspectives direct the speech therapist to new tendencies regarding therapeutic intervention. Aim: to present a bibliographic review of the last two years about chidhood speech apraxia. Conclusion: the present literature about childhood speech apraxia indicates an inclination among the researches that may motivate a mixed therapeutic broaching, in which one contemplates not only aspects pertinent to language processing in a phonological representational level, but alslo aspects related to motor programming and pre-articulatory sequencing of speech itself.
Key Words: Apraxia; Speech; Language.

Resumo
Tema: o aparecimento da desordem práxica durante os primeiros anos do desenvolvimento infantil tem sido freqüentemente denominado de apraxia da fala na infância. As perspectivas teóricas atuais direcionam o fonoaudiólogo a novas tendências em relação à intervenção terapêutica. Objetivo: realizar revisão bibliográfica sobre a apraxia da fala na infância nos últimos anos. Conclusão: constata-se nos textos atuais sobre apraxia de fala na infância uma tendência das pesquisas que poderá desencadear uma abordagem terapêutica mista, na qual se contemple tanto os aspectos pertinentes ao processamento de linguagem num nível representacional fonológico, quanto os aspectos pertencentes ao nível de programação motora e de seqüenciamento pré-articulatório da fala propriamente dita.
Palavras-Chave: Apraxias; Fala; Linguagem.

Recebido em 18.05.2008.
Revisado em 26.07.2008; 24.08.2008; 07.11.08.
Aceito para Publicação em 03.02.2009.

Referenciar este material como:
Souza TNU, Payão LMC, Costa RCC. Childhood speech apraxia in focus: theoretical perspectives and present tendencies (original title: Apraxia da fala na infância em foco: perspectivas teóricas e tendências atuais). Pró-Fono Revista de Atualização Científica. 2009 jan-mar;21(1):75-80.

Introduction

The speech development in children occurs gradually, respecting the stages of maturation and therefore develops trough a large portion of childhood. It is known that children were not born with the speech movements already developed and therefore do not present developed praxis.

The movements of lips, tongue and jaw undergo changes, and undifferentiated movements in early infancy begins to refine and diverse in the development course. These transformations are also fundamental to achieving higher levels of articulatory precision and coordination, which are important to the effectiveness of oral communication (1).

The articulatory gestures of upper and lower lips, and jaw show significant changes during the first years of life and continue its refinement until after 6 years of age (2). The development of motor control of these articulators follows a non uniform course, with jaw preceding the lip. This process of improving the oral motor control significantly influences the acquisition of speech sounds (3).

When this refinement does not occur, the production of speech is compromised, the suspicion of a praxical disorder in childhood may arise. This disorder in childhood is defined as a supposed diagnostic category assigned to children, whose errors of speech differ from the errors of children with delays in the development of speech and are similar to the errors of adults with acquired apraxia(4).

Patients with acquired apraxia of speech usually show alterations primarily in articulation with alterations in the sequence of muscle movements for the voluntary production of phonemes and, secondarily, by prosody alterations, characterized by a slower than normal speech and with a shortage of intonation, rhythm and melody patterns (5). The prosodic abnormalities are usually perceived as secondary to the articulatory difficulties. The lack of speech fluency is caused by pauses and hesitations that occur in an attempt to produce words correctly .The lack of fluency emerges as a compensation for the continuing difficulty in articulating (6).

The onset of the praxical disorder during the early years of childhood development has often been called Developmental Apraxia of Speech (DAS) (4), Developmental Verbal Dyspraxia (DVD) (7) or Childhood Apraxia of Speech (CAS) (8).

From the literature review on the scientific fundamentals in apraxia in childhood published since 1995 the American Speech-Language-Hearing Association (ASHA), Ad Hoc Committee on Apraxia of Speech in Children, adopted in 2007 the term CAS to refer to all apraxia that are manifested in childhood. CAS involves and unifies the studies as well as the evaluation and treatment procedures of all presentations of childhood apraxia of speech.

The designation CAS is, therefore, recommended as the best alternative among DAS and DVD for this speech disorder. The clinical contexts of DVD and DAS are, therefore, incorporated to CAS (9).

CAS is defined as a neurological disorder of speech sounds in childhood. In this disorder, the accuracy and consistency of movements that permeate the speech are hampered in the absence of neuro-mucular deficits. The main obstacle is manifested in the planning and / or scheduling of space-temporal parameters of movements sequences, resulting in errors in the production of speech sounds and prosody (9).

The apraxia in children may indicate three different clinical contexts:

1. It may be associated with a known neurological etiology - intra-uterine disease, infections and trauma.
2. It may occur as primary or secondary signs in children with complex neural-behavioral disorders - genetic, metabolic.
3. It may not be associated with any known neurological or complex neural-behavioral disorder, but yes related to a change on speech sounds of unknown neurogenic origin. These three clinical contexts compose the CAS, updated terminology used for such speech disorder in children (9).

It is estimated that the prevalence of this disorder in the population is 1-2 per 1000 (10), and that the average ratio between boys and girls is as high as 9:1 (7). Although CAS occurs more frequently in boys than in girls, when it is presented on girls, its expression is always more severe (11).

Among the estimated population of 2.5% of pre-school children who present phonological alterations of unknown origin, a ratio of 5% of those children represents children with CAS. These data results estimates that 0,125% of children from general population present apraxia (10).

A study found that 9 in 11 children with CAS (age range from 6.2 to 7.9) present - in addition to motor impairment in speech programming -

language alterations, and also that 55% of these children have family members with Speech and Language alterations (12).

In order to verify the family relationship in CAS, children with suspected CAS and their relatives were subjected to questionnaires and oral expression and motor skills tests. The results showed that the presence of changes in speech and language was higher in families of children diagnosed with CAS than in families of children with other disorders. The family relationship of speech sounds and language alterations was demonstrated in 86% of cases in which at least one family member had been affected. (13).

General characteristics

In the pre-verbal period, children with CAS are described as quiet babies because they tend not to engage in voice playing and their early emissions tend not to develop into different sounds. The fact that babies are quiet signs to the possibility of CAS diagnosis (14).

The period for first significant words emission in children with CAS can happen from 19 months to 4 years of age. The average age for the combination of the first words into sentences of two words also lags behind, appearing between the ages of 33 months and 7 years (15).

A child with CAS can be described as one that shows normal hearing, uses facial expressions, gestures, sounds non-verbal, isolated words or phrases with social communicative intent. These children do not present apparent structural abnormality or paralysis of the oral mechanism, which could justify the absence of intelligible speech. These are children whose skills of listening comprehension seem appropriate for the development of oral communication. However, they are known by the slow progress in therapeutic interventions (16).

A reduction in the rhythm, with syllable segregation, associated with the isochronous or similar time perception, syllables and words not segregated or not co-articulated, considered as prevalent reports in the cases of apraxia of speech is observed in the speech of these children. The rhythmic deficit can be highly specific when compared to the deficit in accent, which may prevail in other types of communicative disorders(17).

Children with CAS have considerable difficulty in the speech production and phonetic accuracy, characterized by slowness, intermittency and variability, acoustically represented by general long-term duration or in segments. Slow speed rate, variable and long pauses (breaks in the speech) and inconsistency among productions are also observed (18). The speech of patients with apraxia presents a narrow range of stress variation in terms of word at sentence level, in addition to the prevalence of nasopharyngeal resonance (19).

The comparative study of prosodic and segmental profiles among on a group of 14 children with suspected CAS and on another group of 73 children with delayed speech of unknown origin showed that the only linguistic domain that differentiated the groups was the presence of inappropriate stress in words and sentences, featuring a pattern of loss of prosodic contrast. The findings also support the heterogeneity of the prosodic profiles associated with apraxia in childhood. Moreover, one concludes that the deficit of stress in this type of apraxia can be more commonly found at levels of phonological representation than at the sequencing of pre-articulation, a characteristic of a speech motor deficit (20,21).

The analytical procedures based on phonology metric and on the use of acoustic measures were shown to be relevant in assessing the search for metric analysis processed by children with CAS, in which a pattern of inappropriate stress sentence was observed. This characteristic stress pattern in childhood apraxia has been proposed as a diagnostic marker for other changes of speech in children. The standard lexical metric found in this review did not differentiate the two groups of children evaluated - language delay and children with apraxia.

However, the study noted that the omission of syllables persisted on a much later age in children with CAS, especially in those who had already been identified as presenting inappropriate sentence stress (22).

Despite the deficit in motor programming of the articulatory structures be shown as the most important deficit in CAS, children rarely experience problems only in speech production. Many of the children have as a consequence or not from the apraxia of speech, a delay in language development, which can be manifested in all linguistic domains, including written language (14).

The literature also describes studies on differences in speech/language and written language among children with suspected CAS and children with other difficulties in speech sounds in school age. Ten children clinically diagnosed with CAS were evaluated from pre-school to school age.

These children were compared to children with isolated alterations in speech sounds (n = 15), and to children with alterations in the sounds of speech and language (n = 14) (8).

Group comparison showed that in pre-school age, the group of children with CAS was similar to the group of children with speech sounds and language alteration. However, at school age, the group of children with changes in speech sounds and language made more positive changes towards learning the language than the CAS group. The study also found that all children with CAS demonstrated commitments of persistent receptive and expressive language. However, the receptive language skills were consistently higher than those of expressive language skills (8).

It is also important to emphasize that there is coexistence of educational problems. These problems are usually presented as a reflection of the difficulties on establishing the necessary relationships between the written representation of words and internal representation that children have of the spoken word. These difficulties bring repercussions to other language skills. The lack of experience in the level of speech production may undermine the experience of reading and reduce proficiency in reading and writing (14).

A variety in characteristics was detected in a study conducted with 75 speech pathologists to verify the criteria used on the diagnosis of CAS. Of the 50 events listed, only six occurred in 51.5% of the responses given by the participants of the study: inconsistent production, oral motor difficulties, hesitation to speak, inability to imitate sounds, difficulty increase proportional to the increase in sentence length and difficulty in producing sounds in sequence (23).

The diversity of characteristics involved in childhood apraxia, even today it is common for children with CAS to be misdiagnosed on phonological disorders or delays in language. Indeed, Speech-Language Pathologists experts in the area tend to associate CAS to an inexplicable lack of progress in treatment. Therefore, there are two characteristics of this disorder for which there is apparent consensus on the literature: the onset occurs early in the child development and presents a course of long-term normalization (4).

Investigations have been conducted with the aim of finding a diagnostic marker for CAS, which would bring implications for both research and clinical practice. This investigations raise the possibility that CAS may be a genetically transmitted speech disorder(4).

Genetic research has been conducted in order to investigate the source of CAS, and among these ones, studies of FOXP2 stand out (19).

More widely spread studies on FOXP2 are the ones carried through with a family entitled as 'KE family'. Half of the members of that family has a phenotype largely marked by an orofacial apraxia; all affected members reported having speech apraxia, some presented language disorders, while others had their verbal performance declined or presented other disorders, and all affected members have a point of mutation on FOXP2 gene (24,25).

FOXP2 is the first gene known to be involved in the development of speech and language. It is located on human chromosome 7q31, and is widely expressed in bilateral neural development (cortex, basal ganglia, thalamus, cerebellum), in areas associated with sensory and motor processes (25).

The studies of structural and functional images provide crucial information in understanding the chain of events by which a point mutation on FOXP2 results in speech and language disorders demonstrated by the KE family. Such studies indicate that FOXP2 may be important for the development of brain networks that are involved in orofacial learning, planning and execution, particularly in motor sequences for speech, as well as conducting manual and other motor sequences (26).

Until a biomarker becomes available for identification of children Who truly present CAS, temporary proposals including diagnosis criteria continue to appear, composing the commonly designated checklist (27).

Researchers use as selection criterion for children with CAS features that include difficulties in sequencing syllables and phonemes, prosodic disorders, deletion of consonants, decreased diadococinese ability, trial and error behavioral, increase in errors on polysyllabic words, inconsistency in articulation with non-usual errors in consonants and vowels (8).

The most recent studies state the importance of the establishment of a diagnostic marker for CAS that considers the speech processing occurring in different stages. There are six stages involved in language processing, which occur in three basic areas:

Input, Output and Organization, which illustrate the possible alternative locations for the production of speech deficits in children with suspected CAS (4):

1. In input process, stages of processing auditory-temporal and memory-perceptual necessaries to the phonological acquisition of the language of the environment are included.
2. In organization process, a stage that reflects the

primitive supra-segmental and segmental of underlying forms, and a transformational stage that adjusts the forms for details underlying morpho-phonemes, allophones and socio appropriate are included. Some theoretical schemes consider that these two levels of phonological processing represent the phonological knowledge of the talker. 3. In the output process two levels are included: one for selecting elements of phonological-recovery and another for pre-articulation sequencing. The final stage of articulatory execution adds any deficit on the integrity of the motor mechanism of speech that is product of previous stages.

In Phonological and Cognitive-Linguistic Planning may be involved in one or more of the five stages of input processing and also in organization; while in Speech Phonetics and Motor Programming there is the involvement of the lowest stage of output - pre-sequencing articulation - as well as the final stage of Articulatory Execution. These aspects illustrate the diversity of possibilities regarding the nature and origin of CAS (4).

In a study carried out with 7 participants, which included 3 children with suspected CAS, two hypotheses were raised. In hypothesis 1, CAS would be the combination of linguistic deficit; in hypothesis 2, CAS would be related to the deficit in the central rhythm for the inaccuracies during speech. That study consisted of measuring the duration of the sub-syllabic components - onset, nucleus and coda - using the Praat 4.0.45 - software for acoustic analysis - comparing the effects of intrinsic and extrinsic vocal duration in monosyllabic words (28).

The results of acoustic analysis of these components allowed to discard the possibility that a specific linguistic difficulty in order to manipulate the sub-syllabic components that are the syllables of words, as raised in hypothesis 1. In the discussions and analysis of the study, the deficit was related to central rhythm, i.e. the combination of time inaccuracies in CAS (28).

The acoustic analysis can be useful in the study of apraxia of speech because the speech of individuals with apraxia tends to be slow, intermittent and variable. The spectrographic analysis notes the obvious differences in words length, being it two times longer than in a speaker with normal speech control. The studies of apraxia of speech based on acoustic show variations of VOT (voice onset time) and errors on phonetic patterns (18).

Procedures for acoustic analysis were used to quantify the temporal regularity in the events of speech and pause in seventy-five children selected from three groups. The following criteria was used on the selection: 1) composed of thirty children with normal speech acquisition, ranging from 3 to 6 years old; 2) with thirty children presenting delay in speech, with the age range from 3 to 6 years; 3) consisting of fifteen children with suspected CAS, with ages ranging from 3 to 14 years(17).

The studies interpreted the restriction on the speech time as the most important feature of the apraxical alteration, which defines a developmental form of apraxia of speech. The acoustic techniques adopted in this study became the easiest quantification and measurement of the time length in the speech events and on events of pause during conversation, forging an important feature of CAS (17).

Conclusions

It is important to detail the complex framework of CAS, as well as their characteristics, in a way that the Speech-Language Pathologist treatment is planned on an appropriate manner. The therapy presents itself as one of the most difficult ones in speech and language disorder. It is a rare disorder, usually difficult to be rehabilitated, and generally the therapeutic processes are long.

There is a consensus on the existence of deficit in the motor control of the articulators for the production of speech. Moreover, the presence of some patterns of errors in speech, such as simplification of syllabic structure, elimination of final consonants, deletion of to initial consonant, reduction of consonantal cluster and stop production is also examined in a linguistic-phonological approach.

However, the emphasis in a phonological non-linguistic only perspective presents itself effective in clinical treatments, as the practice of essentially articulatory gestures are not strengthened enough in this therapeutic approach.

In the present studies on CAS, a trend for studies that may trigger a mixed therapeutic approach can be found. This includes both relevant aspects of language processing at representational phonological level aspects pertaining to the level of motor programming and pre - articulation sequencing of speech itself.

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