BRAIN. Broad Research in Artificial Intelligence and Neuroscience
ISSN: 2068-0473 | e-ISSN: 2067-3957
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2020, Volume 11, Issue 3, Sup.1, pages: 182-196 |
https://doi.org/10.18662/brain/11.3Sup1/134

New Insights into CAS Population Profile and Interaction with Autism – a Wide Retrospective Research

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Abstract: *Childhood Apraxia of Speech (CAS) profile is examined in this article to reveal new insights. These insights might have big influence on the clinical aspect of the phenomenon. One of the interesting insights founded was the close relationship between the CAS population and Autism. Since Autism is such a high incidence phenomenon, it implicates to CAS as well.*

A retrospective research was conducted based on 277 entry level evaluations of children diagnosed with CAS or suspected CAS who visited a private clinic between the years 2006-2013. The analysis included speech variables along background and environmental variables. This article is dealing with the non-speech variables that enable us to observe other parts of this phenomenon.

The main findings are within the areas of diagnosis, age group, educational frames, ear infections, gender, development at first year, babbling and age and speech skill. Some of the results were not as expected.

Further discussion regarding every result and implication is included as well as regarding the interaction between CAS and Autism.

Keywords: *childhood apraxia of speech; autism; NSOME; oral motor; phonetics.*

How to cite: Vashdi, E., Avramov, A., Falatov, S., Yi-Chen, H., Pei-Ru, J., & Mamina-Chiriac, P.T. (2020). New Insights into CAS Population Profile and Interaction with Autism – a Wide Retrospective Research. *BRAIN. Broad Research in Artificial Intelligence and Neuroscience, 11*(3), 182-196. https://doi.org/10.18662/brain/11.3Sup1/134

Introduction

Childhood Apraxia of Speech (CAS) is a considerably new phenomenon and we are still learning its features. We can identify some major characteristics of it, but not really understanding the whole picture. A 9 year old boy, diagnosed with Autism, severe CAS and motor apraxia, found the keyboard and after a year or two of practicing, started typing. His written language appeared to be at peers level. He was asked, why can't you talk? It took him a day to come up with the answer – " I know what I need to do but the tongue won't listen to me, it is like there is disconnection between my brain and my mouth". Regarding his poor grapho-motor skills he wrote; "my hand is separate to me; she got her own will". It was very interesting to look at the CAS in-vivo, from the point of view of the child.

The CAS research is not extensive and there is lack of valid and reliable diagnostic tool, as well as prevalence data and factors that will define the phenomenon and distinguish it from other phenomena. Usually the inspection and research of CAS focus on the characteristics of the speech patterns and the ways to deal with that. This article deals with the profile of the CAS population and tries to conclude from that on therapeutic processes. Not much knowledge exists in the literature about the CAS population. This article will present new insights into the CAS population phenomenon in correlation to the Autism Spectrum Disorder (ASD) population using a wide retrospective research.

CAS speech profile characteristics

CAS is a childhood motor-speech disorder. It is different from Apraxia of Speech (AOS) that occurs among adults after a head injury or Cerebral Vascular Accident (American Speech-Language-Hearing Association. 2007). The updated definition of CAS according to the ASHA is:

"Childhood apraxia of speech (CAS) is a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone). CAS may occur as a result of known neurological impairment, in association with complex neurobehavioral disorders of known or unknown origin, or as an idiopathic neurogenic speech sound disorder. The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody"

Levelt, Roelofs & Meyer (1999) distinguish between three different stages of speech disorders and their pathologies, 1) impairments of lexical access to the word form (classic anomia), 2) phonological encoding (post lexical phonological disorder), and 3) phonetic encoding (apraxia of speech). CAS causes deficits in the production of consonants, vowels and the formation of words.

Shriberg et al. (2003) identified segmental and supra-segmental characteristics of CAS. The segmental characteristics include (a) an articulatory struggle (groping) particularly on word onsets, (b) trans positional (metathetic) substitution errors reflecting sequencing constraints on adjacent sounds, (c) marked inconsistencies on repeated tokens of the same word type, (d) proportionally increased sound and syllable deletions relative to overall severity of involvement and (e) proportionally increased vowel/diphthong errors relative to overall severity of involvement. The supra-segmental characteristics include (a) inconsistent realization of stress (i.e. prominence on syllables or words), (b) inconsistent realization of temporal constraints on both speech and pause events and (c) inconsistent oral-nasal gestures underlying the percept of nasopharyngeal resonance.

CAS diagnosis

Shriberg, Paul, Black & Van Santen (2011) define a group of speech sound disorders (SSD) that is divided into 4 subtypes: speech delay (SD), Speech errors (SE), Persistent Speech Disorder (PSD)(9+ years) and Motor Speech Disorder (MSD). Speech Delay (SD) is the Speech Disorders Classification System (SDCS) classification term for 3–9 year-old children with mildly to severely reduced intelligibility due to age-inappropriate speech sound deletions, substitutions, and distortions. Speech Errors (SE) is the SDCS term for 6–9 year-old children whose speech impairment is limited to distortions of one or two English sounds or sound classes. The Persistent Speech Disorder (PSD) is the SDCS term for speech disorders that persist past 9 years of age and for some speakers, for a lifetime. The Motor Speech Disorder (MSD), includes speakers of all ages whose significant intelligibility deficits are associated with motor speech impairment. CAS and Dysarthria are included in this term. So the CAS can be treated as a motor based etiology subtype within the SSD (Morley, 1975; Shriberg, 2010).

Since there is no good gold standard to define CAS, the diagnosis is still a project in -process. There is no agreed and clear diagnosis tool but rather guidelines based on pathological characteristics. The ASHA convention in 2007 set the term CAS in order to create a universal term, yet

in different countries we can find clinicians and researchers using other terms such as *verbal dyspraxia* or *developmental apraxia of speech*. The non-unity of terms and different perceptions of the phenomenon leads to un-unity in diagnosis.

Forrest (2003) brings the Speech Language Pathologists (SLP) definition or perception of CAS. Seventy five SLPs participated in the study and suggest 50 different items for CAS diagnosis. The most common 6 items appeared in 51.5% of the responses and included inconsistent production, general oral-motor difficulties, groping, inability to imitate sounds, increasing difficulty with increased utterance length , and poor sequencing of sounds. A similar research performed in Sweden came with more consensuses about the main features of CAS. The survey included 25 questions and a response of 127 SLPs was recorded. 85% of the participants suggested inconsistent errors as the core feature of the disorder, 82% noticed difficulties with automaticity and 71% difficulties with sequence maintenance (Malmenholt et al., 2012).

A new diagnostic tool for CAS was published recently and is called the Dynamic Evaluation of Motor Speech Skill (DEMSS) (Strand et al., 2014). The authors used a hierarchical agglomerative cluster analysis in order to identify groups of children with similar features of speech patterns. The DEMSS was able to identify the children diagnosed with CAS but not all of them.

Summarizing these findings suggest that there is still no consensus or clear understanding regarding the main characteristics of CAS, hence, no solid diagnosis that can discriminate CAS from other speech disorder is available. In regards to this study, the definition of CAS was based on previous diagnosis by an SLP or on the guidelines of the current literature. Since the level of speech for all the children participated in this study was very low (25.6% score of single sound production, 8.9% could pronounce only CVCV words and 74.7% couldn't pronounce words at all) and most of the time involved oral motor, sensory and breathing control deficits, the CAS definition was given to them.

CAS prevalence

Shriberg et al. (1997) estimated a prevalence of 1-2/1000 based on the proportion of children referred to one university clinic. Yoss (1975) reported a prevalence of 1% using his criteria for suspected CAS, while Morely 1966 reported 1.3% of CAS. Mckinon et al. (2007) checked the prevalence of stuttering, voice and SSD among 10425 children in 36 primary

schools in Sydney, Australia. The SSD includes CAS and Articulation disorder (characterized by substitution, omission or distortion of speech sounds). Although tested separately, the CAS and articulation score combined post-hoc under one SSD score due to difficulties in differential diagnosis. The SSD prevalence was 1.06%. 13 children were found with CAS (0.12%). This study was taken in a regular primary school so it didn't include the children in the special education school of the same cross sectional sample. Hence the prevalence of the SSD should be higher. Tierney et al. (2015) examined the comorbidity of ASD and CAS. They found that 63.6% of the ASD population diagnosed as well with CAS and 36.8% of the children diagnosed with CAS had ASD as well (N=30). Although it is not a big group, these results contradict the work done by Shriberg, Paul, Black & Van Santen (2011) that postulate no comorbidity between CAS and ASD.

The purpose of the study was to examine various characteristics of the CAS population, in order to better understand it towards more accurate intervention.

Method

A retrospective study was conducted analyzing 277 evaluations of children diagnosed with CAS or suspected CAS. The participants contacted a private clinic for speech evaluation on their own will. The data was collected over the years 2006-2013 of children evaluated at the private clinic in Israel. A set of variables based on the VML method assessment was established for the retrospective data collection. The VML evaluation reliability test demonstrated inter-rater agreement of 81% and correlation of 0.79. Each evaluation was examined thoroughly and the developmental and speech data was extracted according to a detailed index. Each variable had a scale of 3-5 points score with a specific definition of each stage for scoring. Evaluations inclusion criteria were: 1. Suspected CAS or CAS diagnosis. 2. Extracting at least 80% of needed data. Data that wasn't clear enough to fit the variables criteria wasn't used. 3. Evaluations in the Hebrew language only. Evaluations were examined regardless of any other conditions.

Subjects

277 evaluations were examined. Gender distribution - 76.6% boys, 23.4% girls. Average age was 4:11 years old. Age range was 1:7 – 19 years old. All subjects came with a previous diagnosis by a certified examiner (SLP, Neurologist or developmental pediatrician). 13.7% were diagnosed with CAS, the rest of them (86.3%) were diagnosed with suspected CAS.

61.15% of them were diagnosed with Autism Spectrum Disorder (ASD) as well. More details related to diagnosis are discussed in the result section.

CAS definition and criteria

Some of the children attended the Yael Center clinic and participated in this research, were diagnosed with CAS by various SLP's. The others had speech sound disorder and were suspected for CAS. When retrospectively testing the data of each child considering the major characteristics of CAS described in the litrature, all the children fit the CAS guidelines. The average score percentage of single sound production (SSP) was 25.6%. On the words level, 74.7% could not pronounce words in any structure, 8.9% could only pronounce CVCV word structure, 4.4% could pronounce CVCV +CVC word structures, 2.2% could pronounce CVCV +CVC + CVCVC word structures and 8.7% could pronounce more complex word structures accurately based on the mastered SSP. These findings suggest that the basic speech level of the children participated in this study was very low. Based on that and other accepted guidelines we can argue comfortably that they are suspected for CAS.

Measurements

We measured age, diagnosis, speech skill based on the VML evaluation form, educational frame, development during first year, babbling and otitis media effusion.

Results

Diagnosis

The most common diagnosis among the children who came for a speech evaluation was ASD (170 cases, 61.15%). Only 15 of them (8.8%) were diagnosed with CAS as well. Another 6 of them (3.5%) were diagnosed in addition with other syndromes or deficits such as FAPA, Prader willi syndrome, tumor induced epilepsy, Anotia, micro cephalic brain, and ataxia. Only 23 children (8.3%) were diagnosed with CAS only. A big group of children (30.32%) had variety of syndromes and conditions. The children participated in the research came for a speech evaluation, hence had as speech problem. The high incidence of ASD children can't tell us about the prevalence of CAS within ASD but can point of a significant involvement of motor speech deficits among children with ASD.

The results show high incidence of comorbidity (70.6% of all cases). Shriberg et al. (2011) argued that comorbidity of CAS and ASD doesn't exist. In that research the participants had intelligible speech hence reduces the chances for CAS existence. The speech level for the group participated in this research was very low and the incidence of ASD high. These differences in results might suggest of two different groups; ASD with intelligible speech that doesn't show CAS characteristics and ASD with low speech skills that might be related to CAS. The percentage of children diagnosed with ASD and suspected CAS in this study (70.6%) is very similar to the 63.6% founded in Tierney's work and supports their findings (Tierney et al., 2015).

The fact that only 13.7% of the children were diagnosed with CAS regardless of comorbidity with other syndromes points of a serious diagnosis problem, since the different examiners missed a main factor that contributed to the overall phenomenon. The reasons for that might be: 1) difficulties in differential diagnosis, 2) Lack of knowledge regarding CAS syndrome and diagnosis in Israel, 3) Lack of a formal CAS diagnostic tool. If over 60% of children diagnosed with ASD have CAS it means that the population of the CAS might be huge. The ASD prevalence at 2016 in the US was 1:54 (Maenner et al., 2020). It means that the CAS prevalence would be at least 1:111. That prevalence of phenomena requires a special approach from the governmental health agencies. It might mean special education frames and specialists to deal with the motor speech problem.

Another aspect of it is the influence of the motor speech problem on the ASD phenomena. Inability to speak can manifest different symptoms and defiantly will cause a deficit in communication. The contribution weight of the CAS deficit to the ASD diagnosis is not clear but might be crucial. It might also direct the treatment into different primary avenues.

Age of attendance

The average attendance age was 4;11 years (range 1;6 – 17 years). We know that early intervention brings better results (Al Otaiba et al., 2009). An average age of almost 5 years old for an entry evaluation in CAS cases is high. We would rather have an average of 3-4 years old were we can intervene easily with much more time to make a significant change. The implications of the high average age attendance can be:

1) Emotional development - The child has at least 4 years' experience of failure regarding speech. His motivation might be low and so his compliance to treatment. He doesn't believe in himself for making a change.

2) Families motivation – as the child's negative experience, the families might have the same negative feelings about the speech improving chances hence will not be fully motivated. However, they approached and asked for the treatment, hence have some motivation for entering the process. It is much easier to start the journey with a young child and highly motivated family.

3) Level of severity – the older the child is the more severe his problem might be. The results of the age and speech skill correlation doesn't support it fully (see Age and Speech skill correlation), however, we can assume that the child had time to learn and progress and didn't do it and that implicate on the level of severity.

4) Entering school – At the age of 6 years old children move from kinder to school in Israel. The level of speech can sometimes determine the school frame the child will enter regardless of his cognitive level. If the child with CAS starts the special treatment at the age of 5 he is got only one year to get to an acceptable intelligibility for the school system. Often it is not enough time hence speech treatment for a child diagnosed with severe CAS is a long treatment that can last 2-3 years. This point is crucial. In many cases we can observe good cognitive and language skills while the speech is severely damaged. We also know that if we will promote the speech level to the intelligible one word level, the functional change will be extraordinary and might allow the child to go to a regular class. Sometimes starting at the age of 5 is too late and child potential will not be fulfilled due to speech problem and he will enter a special education school.

There might be several reasons for the age attendance problem:

1) Parents or the professional consulting environment Unawareness of the importance of early intervention in those cases.

2) Clinic features – since it is a private, young and unordinary clinic, families might look for more public and mainstream solutions and as second or third option will address this specific clinic.

Age groups

At the very young age (1-2 years) 6 children attended the clinic while the number raised to 55 children at the next age group (2-3 years old) and stayed at the same level for the next 3 age groups. At group age 6-7 years old the number of children started to drop, and that trend went on for the next age groups. The fluctuations in group sizes might result from the following explanations:

1) It demonstrates the families need for the treatment. In the first group of the very early age the families are not convinced that there is a problem and rather wait with the treatment. In the ages 2-6, the speech problem greatly effects communication and social skills so the need for the intervention is the highest. After the age of 6, the belief of a possible change in speech reduces so the group sizes reduce as well.

2) The evidence of the speech problem – in the very early age the speech problem is not so acute hence not as evident. In the ages 2-6 years old, the speech problem is more acute and requires the treatment hence more children will come to have the treatment. For the group ages of 6 + there might be less children needed the treatment since they have been treated already.

3) Education system structure in Israel – from early age children with special needs attend special education kinder in Israel until the age of 6. At that age they move to special education schools. The school timetable is more rigid then at kinder and the child usually will spend more time in the school. It is harder to have a secondary treatment schedule in school setting, in comparison to kinder and that might be another reason for the change in group sizes from the age of 6.

Gender

The gender distribution for most of the developmental childhood syndromes involves higher rates for boys than girls. Nicholas et al., (2009) found a male to female ratio of 4.7:1 in an ASD population of 8156 4 year-old. Fernell & Gillberg (2010) report boys to girls ratio of 5.1:1 in an ASD population. Boyle et al. (2011) conducted a wide research to explore trends in prevalence of developmental disabilities in the US. They have found a boys to girls overall ratio of 1.89:1 and 3.89:1 in the ASD population. In this study, 77.4% of the children were boys and 22.6% girls. A ration of 3.4:1 boys to girls. These findings are concurrent with previous literature reports. The girls age was significantly higher than the boys (5.73 and 4.68 respectively, T test = 0.0029). On the other hand, The SSP scores for boys and girls didn't differ (26.08% and 24.18% respectively, T-test= 0.66), hence, gender had no effect on the severity of the symptoms.

Educational frame

In 17 cases there were no data regarding the educational frame. Half of the children went to a communication therapy oriented educational frame (N=132, 50.76%). 88% of them were ASD. 20% (N=52) went to regular schools while a similar group (N=51, 19.61%) went to other special

education frames. Only 1.53% (N=4) attended small classes in regular schools. An interesting result was the amount of children in home schooling (N=21, 8.07%). For CAS as a pronunciation deficit there is no specific educational frame that will support the pronunciation severe needs. If the child has a specific syndrome then he will be placed in the matched educational frame but the focus of that frame will not be speech by definition. A communication kinder will focus on communication skills, Language kinder will focus on language skills etc. The children with pure CAS have no matched educational frame that will answer their needs.

Most of the children were diagnosed with ASD hence naturally were placed in a communication kinder or school. It is interesting to see that 12.12% of the children attended communication frames didn't have ASD. It means that children without communication problem but severe pronunciation deficits attended an educational frame that couldn't support them. The explanations for that might be: 1) No other educational frame in their leaving area, 2) it is a very good educational frame that the parents believed will promote the child.

Development during first year

In 20 cases there wasn't enough information to assess development. In 92 cases normal development was recorded (35.93%) with average SSP of 28.86% and age average of 4;3 years. In 34 cases delayed development was recorded (13.28%) with average SSP of 23.74% and age average of 3;8 years. In 130 cases severe delayed development was recorded (50.78%) with average SSP of 20.44% and age average of 5;5 years. The results show a tendency towards significant difference in SSP between groups (alpha=0.1), hence there might be a relationship between the general development during the first year of life and speech level among children with CAS. The group of severe developmental delay showed the lowest speech skill even though the age average was the highest. It is not clear why general development will affect the speech acquisition. A possible explanation might be the amount of attention that the child needs to put in different areas might sometimes take the focus of other areas hence delay their development. For example, a delay in walking might delay other areas such as speech since the child is still focusing on the motor aspect of development and having control over his body.

Babbling

Babbling is an early stage of verbal communication. It is a communication and motor phase in the first year towards first syllables and

words. The absence of babbling might be a precursor for further development (Burlea et al., 2010). We had data for scoring babbling in 187 cases (67.5%) on a 1-3 scale. The first was typical babbling during first year, the second was reduced babbling, and the third was no babbling at all. In the normal babbling group were 76 cases (40.6%) with average SSP of 26.28%. In the reduced babbling group were 35 cases (18.7%) with average SSP of 22.88%. In the no babbling group were 76 cases (40.6%) with average SSP of 12.19%. The results indicate that no babbling during the first year is related with lower speech skill later and can point of more severe CAS condition. One way ANOVA test demonstrated significant differences only between the normal babbling group and the no babbling group (a=0.007)

Otitis media effusion (OME) and grommets

The ability to pronounce sounds depends among other things on the auditory input. Deafness will result many times with no speech at all, while disturbances in auditory input due to different reasons can affect speech (Lupu et al., 2016; Lupu et al., 2015; Lupu et al., 2016). Ear infection and fluids in the middle ear (OME - otitis media effusion) is an example of that kind of disturbance (Klein 1984; Roberts et al., 2004). It is a question whether an auditory input deficit such as OME should be part of CAS definition or relate to CAS. The ability to plan a motor program relays on the sensory input in the first place (Rose & Christina 2006). In 35 cases there was insufficient data regarding OME and grommets. In 63 cases (25.6%) OME was reported. Only 40 of them (63.4%) were treated with grommets, all with success. 74.4% of all cases were reported of not having ear infection. The blowing scores for the children with and without OME were 3.03 and 2.96 respectively. These scores represent an average ability to blow without an accessory in the mouth but without control of power or direction. No significant difference was found between the groups; hence the OME has no influence on movement control of blowing. The tongue movement control for the children with and without OME was 2.63 and 2.89 respectively. These scores represent an average ability to stick tongue out and in some cases move it to the sides of the mouth. No significant difference was found between the groups; hence the OME has no influence on movement control of the tongue.

The interaction between OME and pre-verbal skills suggests that the children had suspected CAS regardless of the OME, since the occurrence of the oral motor deficit wasn't related to OME. OME prevalence was 25.6% which is similar to the prevalence in typical developed children without CAS (Paterson et al., 2006). In comparison, 37% of children with down syndrome

were found candidate for grommets surgery (Barr et al., 2011). It seems that OME is not a unique factor in CAS however can contribute to the difficulties if other CAS characteristics exist.

Limitations

Since this research is a retrospective data collection and analysis, not all the variables researched existed in all the cases. Some of the data needed to be speculated from the texts causing possible errors in data collection. However, the inter-rater tests showed good agreement. The data represents the population that came for speech therapy in the clinic during 2006-2012 so no random selection of cases was done. However, no selection of cases was made from the attended cases. The results refer to Hebrew speaking children only. We should consider all those limitations when concluding towards clinical purposes and theoretical hypothesizes.

Summary

This research is unique in size regarding the CAS population and shed light on the population's profile. The better understanding of age, gender, diagnosis, development during first year and the other topics, might change the attitude towards treatment. It can affect the onset of treatment, prognosis expectations, inclusion policy and understanding of trends of the phenomenon behavior. The comorbidity of ASD and CAS that is presented in this article promote us to examine carefully the causes of the communication problem of the child diagnosed with ASD, since sometimes the CAS component of it might be crucial. That might lead to different intervention.

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