

CLAUDIO ZMARICH, SIMONA BERNARDINI, SABRINA BONICHINI,
MIRKO BONOTTO, FEDERICA CHIARI, ERIKA GANASSIN,
CATERINA PISCIOTTA, FABIOLA VULLO, ELISA ZACHEO

Linguistic proficiency, communicative attitude, and severity in a sample of children who stutter aged 4-6 years

Children who stutter develop self-awareness of their speech from an early age, resulting in a more negative communication attitude (CA) compared to their fluent peers. Other speech disorders can also affect CA. The relationship between low scores in linguistic proficiency and negative CA among children who stutter without other comorbidities is still unclear. Eighteen children (aged 4;0-6;11) diagnosed with stuttering, but not yet treated, Italian speakers from Veneto, with no other concurrent disorders were assessed using KiddyCAT for CA and BVL_4-12 for linguistic proficiency. Their linguistic abilities were comparable to those of fluent peers, except for their poor performance on articulation and repetition tests. Although most children stutter moderately or severely, their CA was comparable to that of their normal, fluent peers. No significant correlations emerged between CA, language ability, and stuttering severity.

Keywords: children who stutter, Communicative Attitude, stuttering severity, linguistic proficiency, non-words repetition.

1. *Introduction*

Although the International Classification of Diseases has currently arrived at the 11th edition (ICD-WHO, 2021), we still prefer the definition of stuttering contained in the 9th: “disorders in the rhythm of speech, in which the individual knows precisely what he wishes to say, but at the time is unable to say it because of an involuntary, repetitive prolongation or cessation of a sound” (ICD-WHO, 1977, p. 202). Our preference is motivated by the central role assumed here by the “involuntary” character of the stuttering’s disfluencies. One can easily imagine what kind of despair and frustration can be induced by such an experience of involuntary loss of control (Perkins, 1990). These negative feelings take place from the beginnings of stuttering for most of the children who stutter (CWS), and then they give rise to a constellation of reactions which are an integral part of the stuttering syndrome and have received the label of “secondary”, or “associates”, symptoms (Yairi, Seery, 2023).

Secondary symptoms are primarily caused by a negative communication attitude (CA) towards one’s own speech, along with an image of oneself as a poor/bad speaker. In the upcoming exposition, we will address: 1) the concept of CA and its significance for CWS; 2) its applicability to other speech and language disorders,

not just stuttering; 3) the high rate of comorbidity of CWS with speech and language disorders; 4) the speech and language factors interacting with stuttering in early childhood.

1.1 Communicative Attitude and stuttering

The close relationship between stuttering and CA has been the subject of important research and debates in recent decades. To date, clinicians have embraced the idea that stuttering should be considered as a complex and composite disorder. The manifest symptoms, in fact, represent the central, but not the only, attribute of this multifaceted pathology, which also includes the experience of the person who stutters (PWS). According to this perspective, the evaluation and treatment of PWS must be addressed to the disfluency and to the negative emotional reactions, to the secondary dysfunctional coping behaviours attempted by the subject to overcome and/or avoid stuttering, and to the cognitive dimension. The therapeutic process can only be considered positive and satisfactory if verbal disfluency is reduced and the CA is normalized. (Menzies, O'Brian, Onslow & Packman, 2008). In fact, negative CA appears to play a fundamental role in the maintenance of the disorder (Guttormsen, Kefalianos & Næss, 2015; Bloodstein, Bernstein-Ratner & Brundage, 2021).

Research has established that stuttering typically starts in preschool years (Yairi, Seery, 2023) and that children could be aware of the dysfluency about their speech as early as they are three years old, at the same time or shortly after the stuttering onset, and this awareness increases along with age. Hence, CWS not only can be aware of their disfluency from the beginning, but they also develop a perception of their disfluency as negative. This negative speech-related attitude includes viewing themselves as bad communicators, evaluating speech as difficult, being apprehensive in speaking situations and feeling high anxiety when communicating with others. (Ambrose, Yairi, 1994; Blood, Blood, Tellis & Gabel, 2001; Ezrati-Vinacour, Platzky & Yairi, 2001; Mulcahy, Hennessey, Beilby & Byrnes, 2008; Iverach, Rapee, 2014). This can lead CWS to implement waiving and communication delegations to alleviate the anxiety due to the fear of being judged negatively by the interlocutor. Unfortunately, a communication disability such as that caused by stuttering, can have negative repercussions on the overall quality of life of the PWS throughout the lifespan (Yaruss, Quesal, 2004). These include limitations in social interactions, prejudices and social stigma, distress, shame, and low self-esteem (Cream, Onslow, Packman & Llewellyng, 2003; Ginsberg, Wexler 2000). Stuttering sometimes leads PWS to encounter limitations in school career and career choices (Daniels, Gabel & Hughes, 2012; Gabel, Blood, Tellis & Althouse, 2004). Not only CWS but also the children who do not stutter (CWNS) can be aware, as early as when they are three years old, of the stuttering-like-disfluencies (SLD) produced by CWS, and they can give them a negative judgement. As a matter of fact, CA tends to be more negative in CWS compared to CWNS (Guttormsen *et al.*, 2015).

A measure of communication attitudes, such as the *Communication Attitude Test for Preschool and Kindergarten Children Who Stutter* (KiddyCAT, Vanryckeghem, Brutton, 2007), has been shown to meet the criteria of assessing the attitudes towards communication in preschool-age children. Recently, KiddyCAT has been translated and adapted to Italian (Vanryckeghem, Brutton, 2022).

It had been speculated that factors such as age, severity, history of stuttering (e.g., time since onset), gender, parental influences, metalinguistic awareness, treatment and language proficiency may influence awareness of stuttering and negative attitude about speech (Ambrose, Yairi, 1994; Yairi, Seery, 2023; Boey, Van de Heyning, Wuyts, Heylen, Stoop & De Bodt, 2009).

There are contradictory results regarding age. A number of studies found that age did not significantly affect the difference in CA between CWS and CWNS (Vanryckeghem *et al.*, 2005; Bernardini *et al.*, 2009; Bernardini *et al.*, 2019). However, other studies have shown that the CAT scores of CWS tend to increase with age (Kawai *et al.*, 2012), while those of CWNS tend to decrease (Vanryckeghem & Brutton, 1997; Brutton, Vanryckeghem, 2007; Clark, Conture, Frankel & Walden, 2012), leading to the result that the differences in CA between CWS and CWNS increase with age. The review by Guttormsen *et al.* (2015) supports the hypothesis that the CA of CWS worsens with increasing age. The authors suggest that CWS may be exposed to increasingly negative communication experiences as they grow older: for example, they may experience difficulties in school, where communication needs become increasingly demanding, and where they are also at greater risk of being bullied (Blood, Blood, 2004; Cook, Howell, 2014). However, not all studies conclude that CA gets worse with age. For example, in the study by Clark *et al.* (2012) the group of younger children (from 3;0 to 4;6 years) obtained higher scores (more negative attitude) on the KiddyCAT than that of the older ones (from 4;7 to 5;11 years), and Groner *et al.* (2016) also found that CA improved with increasing age in both CWS and CWNS. The age where CWS scores start to differ from those of CWNS has been investigated in research on CA during the developmental period. Thanks to the use of the KiddyCAT, it was found that the CA of the CWS and that of the CWNS begin to differ around the age of 3-4 years, when stuttering usually begins (Bernardini *et al.*, 2009).

As to severity, in adults and adolescents, this is not linearly correlated with either CA or the overall impact of the disorder. Specifically, for some PWS the severity is mild, but the impact of the disorder is high, while for others the opposite may occur (Winters, Byrd, 2021). Conversely, research in school-aged children suggests that there may be a relationship between CA and severity, although there are some conflicting findings. Ambrose, Yairi (1994) did not find any relationship between the severity of stuttering and stuttering speech awareness. More recent studies suggest the existence of such a relationship, as they found that children with more severe overt symptoms had more negative CA (Beilby, Byrnes & Yaruss, 2012; Kawai, Healey, Nagasawa & Vanryckeghem, 2012; Vanryckeghem *et al.*, 2001). However, other studies do not confirm this relationship: Groner *et al.* (2016)

provided evidence both in favour and against it, while in the study by Winters, Byrd (2021), none of the measured characteristics of severity (frequency and duration of disfluencies, physical concomitants) correlated with CA. Given these conflicting results, we cannot conclude that CA is associated with stuttering severity, but neither can we rule out this possibility. If this hypothetical relationship could be confirmed, it would help clinicians to identify early children who need immediate treatment (Guttormsen *et al.*, 2015).

Regarding the relationship between CA and gender, there is consensus on the fact that CAT or KiddyCAT scores do not differ significantly between boys and girls who stutter (Vanryckeghem *et al.*, 2005; Bernardini *et al.*, 2009; Clark *et al.*, 2012), as confirmed by a recent meta-analysis (Guttormsen *et al.*, 2015).

1.2 Communicative Attitude and Speech and Language disorders different from stuttering

Despite the wide debate on the subject, numerous studies support the hypothesis that CWS have a high probability of exhibiting concomitant disorders, especially of a linguistic nature (Blood, Ridenour, Qualls & Hammer, 2003). In the light of the fundamental role played by CA in stuttering, it seems useful to review some of the main evidence in the literature regarding the characteristics assumed by CA in other speech and language disorders.

The study by De Nil, Brutten (1990), using the CAT on children aged 7 to 14 years, allowed to compare the CA of the CWS with that of peers affected by other speech disorders. Participants were divided into four groups based on the disorder they experienced, including stuttering, articulation problems, voice problems, and a control group. The results highlighted the presence of more negative attitudes in all speech disordered groups than in the control group, although the highest scores were obtained by CWS and by children affected by voice disorders (De Nil, Brutten, 1991). Evidence of negative CA has also been reported in 10-year-old children with cleft lip and palate (Havstam, Sandberg & Lohmander, 2011). They scored higher than their normal fluent peers on the Swedish version of the CAT test (CAT-S) (Johannisson, Wennerfeldt, Havstam, Naeslund, Jacobson & Lohmander, 2009).

A study analyzed the CA of a group of children with speech sound disorders (SSD) in school and preschool age using the KiddyCAT test and *The Child Form of the Speech Participation and Activity Assessment for Children* (SPAA – C) (McLeod, 2004; cited in McCormack *et al.*, 2019), an informal questionnaire about how the child perceives his own speech. The results revealed that 58.9 % of the participants achieved a score on the KiddyCAT index of positive CA, while the remaining 41.1 % achieved a score much higher than expected for the corresponding age, thus showing a tendentially negative attitude. Furthermore, by relating the KiddyCAT data to the child's articulatory accuracy, it emerged that the more negative CAs were correlated with lower phono-articulatory skills, expressed by a lower *Percentage of Correct Consonants* (PCC) (McCormack *et al.*, 2019). The study then examined the relationship between the parents' assessment of the child's intelligibility, the

perception they reported of their children's CA, and the attitude that the latter showed in the KiddyCAT. The results indicated a substantial discrepancy between these measures, revealing that the child's CA was not correlated with that perceived by the parent nor with the level of intelligibility reported by them. This represents a further confirmation of how, in clinical practice, it is necessary to adequately take into consideration the point of view of the child, as well as that of the parent (McCormack *et al.*, 2019).

The possibility that disfluency awareness and negative CA could not to be associated in a direct way is suggested in a study by Choi, Hurd & Kim (2022) on CWNS: they found that those with higher language ability are more likely to be aware of, and negatively evaluate, disfluent speech than the children with lower language ability. Perhaps, children with higher, compared to lower, language abilities seem more aware of differences and deviations in speech disfluencies, because they have a better speech language monitoring system to monitor their speech output in terms of phonology, morphology syntax, semantics, and fluency, so as to produce error-free speech (Levelt, 1993). This result suggests that children affected by speech and/or language disorders (like stuttering) may not have a speech-language monitoring system sufficiently developed, which may cause them not to be aware of their problems and consequently not to be negatively affected in their CA.

1.3 Comorbidities of stuttering with speech and language disorders

Current research highlights the presence of concomitant speech and language disorders and/or other disorders in approximately two-thirds of CWS (Blood *et al.*, 2003). However, this figure is affected by numerous differences between the various studies, attributable to the different methodologies used and the composition of the samples, in terms of age of the participants and diagnostic inclusion criteria (Arndt, Healey, 2001).

From a clinical point of view, studies concerning comorbidity with stuttering assume considerable importance, since the evaluation and treatment of children with concomitant disorders has very different connotations compared to those relating to isolated stuttering (Blood *et al.*, 2003).

Furthermore, according to some authors, phonological disorders associated with stuttering in preschool age are a potential negative prognostic indicator of stuttering recovery (Conture, 2001), although there is evidence to the contrary (Watkins, Yairi, & Ambrose, 1999).

Historically, the hypothesis of comorbidity between speech disorders and stuttering refers to the DC Model (Demands and Capacities Model; Adams, 1990; Starkweather, Gottwald, 1990), which argues that the conversation demands can induce an overload of the language production systems of the child and a consequent increase in disfluent episodes when they exceed his motor, linguistic and emotional features. Arndt and Healey (2001) highlighted the presence of associated linguistic disorders in 44 % of 205 CWS aged between 3 and 20 years, with a share of phonological disorders equal to 33 %. Data seems to support the

hypothesis of a higher incidence of phonological disorders in CWS compared to the general childhood population (20 % of associated phonological disorders in CWS compared to 2.6 % among CWNS) (Arndt, Healey, 2001; Blood *et al.*, 2003).

Blood *et al.* (2003) reported the presence of associated linguistic disorders in 62.8 % out of a sample of 2628 CWS, with a percentage of articulation disorders equal to 33 % and 12.7 % of phonological disorders. The latter did not correlate with the level of stuttering severity (Walk, Edwards & Conture, 1993; Yaruss, Conture, 1996). Other linguistic disorders frequently found in CWS concern the lexical-semantic production (13.5 %) and lexical-semantic understanding (12.1 %) (Blood, *et al.*, 2003), as well as voice problems (0.12 %) and SSD (Speech Sounds Disorders), with a prevalence of 1.06 % (McKinnon, McLeod & Reilly, 2007).

In contrast to the research mentioned above, the review by Nippold (2012) and a study by Smith and Weber (2017) suggest that CWS, as a group, are no more prone to language impairment than CWNS, but report heterogeneous linguistic abilities placing themselves on all levels of performance.

1.3.1 Comorbidities of stuttering with disorders different from speech and language disorders

According to Blood *et al.* (2003), 34.3 % of the sample shows concomitant disorders with primary causes outside from the speech and language ability. Specifically, they involve: 15.2 % of cases with associated DSA (8.2 % of which diagnosed with dyslexia and/or dysorthography), and 5.9 % with concomitant ADHD. Among the co-occurring non-language disorders, five were found to have a frequency of less than 5 %. They include: central auditory processing disorders (3.1 %), neuropsychological disorders (2.9 %), behavioural disorders (2.4 %), sensory integration disorders (2.1 %) and acquired disorders (1.3 %). The incidence of concomitant disorders (both linguistic and non-linguistic) affects males more than females, especially for concomitant speech-sound disorders (48.2 % of males against 38.8 % of females) and learning disorders (12.2 % of boys versus 8.3 % of girls) (Blood *et al.*, 2003).

The systematic analysis of health information by the electronic health record (EHR) provided a more comprehensive view of the comorbidity between stuttering and other disorders: it compares 572 subjects diagnosed with developmental stuttering with 2765 normal fluent subjects. The aim of the study was to verify the possibility of identifying possible disease associations existing with stuttering, using a study approach known as *PhеWAS* (Phenome-Wide Association Study), that uses *Phеcode* (labels inspired by the ICD 9 classification), including all the phenotypic information with common characteristics. In this regard, associations were found with Neurodevelopmental Disorders and Language Disorders, followed by Pervasive Developmental Disorders and ICT (Pruett, Shaw, Chen, Petty, Polikowsky, Kraft, Jones & Below, 2021). Autism spectrum disorder does not appear to be in statistically significant comorbidity with stuttering, while the strong co-presence of deafness and stuttering is in contrast with most of the evidence in the literature, reporting a low prevalence of hearing loss in the stuttering population (Montgomery, Fitch, 1988; Yairi, Seery, 2015).

1.4 Speech and language factors impacting stuttering in childhood

There are many ways to evaluate speech and language proficiency in CWS with respect to CWNS. The first distinction to be made is between behavioural and imaging measures. Due to space restrictions, we will give prominence to the behavioural studies. The reader interested in brain imaging and electrophysiological studies can refer to Chang *et al.* (2018) and Chow *et al.* (2023), among others. Within the behavioural studies, we can further distinguish among studies applying standardised/commercial tests (or batteries) to samples of CWS and CWNS (community-screening studies included), and studies using nonstandard experimental methodologies to assess one or more specific capacities of CWS.

1.4.1 Standardised test batteries

The study by Coulter, Anderson & Conture (2009), conducted using various standardised tests for the evaluation of the linguistic abilities of CWS and CWNS, showed the CWS group to be higher than the norm as to general linguistic abilities, but lower than the norm as to those more frankly phonetic-phonological. The research followed the model of the study by Anderson, Pellowski & Conture, (2005), according to which the CWS have, on average, three times the probability of showing dissociations between the different language skills compared to CWNS. A meta-analysis of 22 studies investigating the language skills of CWS and CWNS aged 2-8 years also revealed subtle differences between these two groups in measures of general language (MLU), and receptive and expressive vocabulary. The performance of the CWS, although within the norm values, was in fact poorer on average than that exhibited by CWNS (Ntourou, Conture & Lipsey, 2011). It has been hypothesised that the frequent dissociations between the single linguistic levels found in CWS could make their linguistic processing system more vulnerable to interference on the cognitive, sensory-motor and emotional level during conversation, increasing the likelihood of experiencing disfluent episodes (Bosshardt, 2006; Coulter, Anderson & Conture, 2009; Clark, Conture, Walden & Lambert, 2015; Smith, Weber, 2017).

Numerous studies have placed CWS into different subgroups based on the language skills they exhibit (Yairi, 2007; Hubbard-Seery, Watkins, Mangelsdorf & Shigeto, 2007; Reilly *et al.*, 2009). A first subgroup includes CWS with poor language skills and high comorbidity for language disorders (Conture, Zackheim, Anderson & Pellowski, 2004; Hollister *et al.*, 2017; Walden *et al.*, 2012), while a second subgroup includes CWS who obtain better scores than CWNS in the main standardised tests for the assessment of linguistic skills and in all measures of expressive language (Watkins, Yairi & Ambrose, 2005; Watts *et al.*, 2017).

These conclusions have been criticised by Nippold (2012; 2019), as many of the studies included by Ntourou, Conture & Lipsey (2011) were not adequately balanced in the sample recruitment criteria, the socioeconomic characteristics of the participants (SES) and the different language tests administered. Nippold's meta-analysis reported opposite conclusions to those of Ntourou and collaborators,

highlighting how CWS show linguistic skills equal or superior to those exhibited by CWNS of the same age, although these cannot be defined as exempted from linguistic weaknesses (Nippold, 2012; 2019). Indeed, there is evidence demonstrating the presence of impaired language skills in some CWSs (Smith *et al.*, 2012), like what can be found in some CWNSs (Leonard, 2014). Perhaps the most balanced and convenient description has been put forward by Smith, Weber (2017): CWS as a group would obtain highly heterogeneous performances, with higher percentages of individuals ranking at an extremely good or extremely poor level (Yaruss, LaSalle & Conture, 1998; Hubbard-Seery *et al.*, 2007).

Despite the lack of evidence suggesting that CWS are truly language-disordered, numerous studies have demonstrated subclinical impairment of language skills in cohorts of CWS. Two relatively recent community-screening studies (Zaretsky *et al.*, 2017; Shimada *et al.*, 2018), were able to compare language skills in CWS with much larger cohorts of CWNS in the same jurisdiction, all of whom were screened as part of public health projects. Both found relative depression of oral language skill in CWS, especially regarding articulation and grammar.

In contrast, an Australian community health project, the Early Language in Victoria Study (ELVS), using different screening methods, found a slight oral language advantage, rather than disadvantage, for CWS that they tracked, compared with children who were never reported as stutterers (Kefalianos *et al.*, 2017). However, one large-scale study of 100 pairs of CWS and CWNS collected across numerous American laboratories (Luckman *et al.*, 2020) found that CWS scored within 1 SD lower in both expressive and receptive vocabulary, as measured by standardised assessments, than did appropriately matched fluent peers.

1.4.2 Phonetic and phonological levels

According to Sasisekaran (2014), it is possible to distinguish at least three different points of view on the role of Phonology in stuttering (she does not consider the level of articulatory planning and delivery):

1. the study of the phonological structures that predict the placement of SLDs in the speech chain;
2. the exploration of phonological competence through standardised tests;
3. the study of cognitive processes related to phonological processing (retrieval and construction of a phonetic plan from the lemmas to the utterance);

As to 1), in the words of Brundage, Bernstein-Ratner (2022: 8), “adult speakers are more likely to stutter on words beginning with consonants, on longer words, on content words, and on words occurring early in the utterance. Long and complex utterances also increase stuttering frequency when compared with short, simple utterances”, and CWS differ from adults only for the tendency to stutter on function words (Howell, Au-Yeung, 1995; see also Bloodstein, Ratner & Brundage, 2021). The literature has often explored the link between the articulatory complexity of the word to be produced and the likelihood of encountering disfluent episodes., noting for instance how CWS usually make less use of complex consonants than CWNS (Howell, Au-Yeug, 1995) and how SLDs occur more frequently in

word-initial position, and more in stressed than in unstressed syllables (Howell, Au-Yeung & Sackin, 2000; Natke, Sandrieser, Van Ark, Pietrowsky & Kalveram, 2004). Various studies also report the presence of subtle differences in motor aspects between the speech of CWS and that of CWNS. These differences include laryngeal function (reduced amplitude and intonation range in phonation), a lower amplitude of articulatory movements and a lower speed of speech (Yairi, Ambrose, 2005). The authors support the hypothesis of poor motor coordination stability and less flexibility in speech planning in preschool CWS: the speech of these subjects appears in fact more vulnerable to variations in the speech rate, linguistic complexity, and social and cultural pressure (Smith, Zelaznik, 2004; Walsh, Mettel & Smith, 2015).

These data have broad clinical utility, as they allow early prediction of the probability of remission or persistence of the disorder. According to literature, CWS who have better articulatory skills are more likely to experience spontaneous recovery than those who have lower scores on linguistic articulation tests (Spencer, Weber-Fox, 2014). Conversely, CWS presenting an excessive degree of intrasyllabic coarticulation for a period longer than the 6th/7th month from the onset, would more frequently encounter the persistence of the disorder (Subramanian, Yairi & Amir, 2003).

The twenty-year-long research project by Yairi and colleagues has also shown that CWS, two years after the onset, are on average later than CWNS in phonological development (Paden, 1996). Notably, some authors have reported a more diverse typology of phonological processes in the speech production of preschool CWS than in CWNS. These processes are often age-inappropriate or atypical, although they are rarely categorised as a real disorder (Louko, Edwards & Conture, 1990).

The hypothesis of a greater immaturity in the phonological processing of preschool CWS was instead supported by the research of Byrd, Conture & Ohde (2007), related to the facilitating effect (priming) of lexical access following holistic acoustic stimuli (all the disyllabic word minus the initial consonant) and incremental (only the initial consonant and the transition to the next vowel). These studies have shown how the first type of elaboration, typical of early developmental stages, facilitates the recovery of the lexical target in children with and without stuttering already at the age of 3, while the second, typical of the adult population, appears to be only advantageous for CWNS ages 5 and up.

According to psycholinguistic theory, the disfluencies would also be caused by a phonological coding deficit, related to the recovery and construction of the phonological segments of words (Pelczarski, Yaruss, 2016). A fundamental role in this regard is played by the phonological memory, which allows the sound representation to be formed and preserved for a sufficiently long time to be correctly reproduced (Pelczarski, Yaruss, 2016). It is usually tested by non-word repetition trials, in which CWS perform on average worse than CWNS (Hakim, Bernstein-Ratner, 2004; Anderson *et al.*, 2006; Anderson, Wagovich & Brown, 2019). The difference assumes significance especially for the CWS under the age

of 4-5 years with a greater risk of becoming persistent (Bankson, Bernthal, 1990), and is detected more frequently on trisyllabic words, like what is evidenced in the population with a diagnosis of DPL (formerly DSL) (Hakim, Bernstein-Ratner, 2004). Furthermore, as with DPL, the phonological memory impairment found in CWS may have cascading effects on lexical and syntactic development, limiting the acquisition and retention of new words and their subsequent organisation into sentences (Coady, Evans, 2008).

1.4.3 Syntactic and morphological levels

The influence exerted by morphological and syntactic variables on the probability of incurring disfluent episodes is now known in the literature. In particular, in the CWS population, disfluencies have been shown to occur more frequently in sentence-initiating words (Richels *et al.*, 2010; Buhr, Zebrowski, 2009), and in function words rather than content words (Howell, Au-Yeung, & Sackin 2000). Furthermore, the onset of the disorder occurs when the acquisition of the syntactic ability to produce the utterance is developing, between the ages of 2 and 4 years. In this stage, the planning of the utterance switches from a purely semantic level to a syntactic one, with a discontinuity that can be translated into an increase in the frequency of disfluencies and self-corrections (Wijnen, 1990). The latter, according to some authors, could be associated with the excessive demand exerted by the growing syntactic complexity on a still underdeveloped linguistic system (Zackheim, Conture, 2003).

One of the most frequently used measures for the indirect evaluation of the child's syntactic development is the Mean Length of the Utterance (MLU), typically calculated, in Italian, as the ratio between the total number of words produced and the total number of utterances (Brown, 1973). This tool becomes unreliable in capturing the internal syntactic complexity of the utterance produced by the child when he acquires the ability to combine 3 elements in a sentence (approximately 3 years of age, Pinton, Lena, 2015).

According to the aforementioned meta-analytic review carried out by Ntourou and colleagues (2011), the spontaneous speech of CWS would see the production of on average shorter utterances (poor MLU), compared to those of CWNS. This difference may be attributable to the possible strategies for avoiding disfluencies implemented by CWS, or to their temperamental aspects (among the characteristic factors responsible for the reduction of verbal output, there would be, for example, the scarce propensity to report in an evaluation context).

1.4.4 Lexical-semantic level

Lexical skills allow to learn and distinguish the single words of a vocabulary, while semantic skills link these words to related representational and operational concepts. In the child's linguistic development, the lexical and semantic aspects are strongly connected, as initially the subject tends to produce words with an excessively generalised meaning (holophrases) and enriches his vocabulary only after repeated experiences of the world around him (Deodatus, 2014). To accurately assess these

skills, it is important to take into account the personal and social variables that affect the child's language exposure and experiences. (Luckman, Wagovich, Weber, Brown, Chang, Halle & Bernstein Ratner, 2020). It is in fact known that lexical skills tend to increase with age, are more developed in the female population at an early age (Eriksson, 2012; Luckman *et al.*, 2020), and are positively influenced by a high familiar SES (Hoff, Tian, 2005).

With regard to stuttering, many studies have reported heterogeneous results regarding the lexical skills of CWS compared to those of CWNS. Such different results could be attributable to the methodological differences found in the various studies differing on the abovementioned variables (age, gender, SES).

Among the studies that have found a poorer performance in CWS compared to CWNS there is that of Silverman, Bernstein-Ratner (2002), aimed at assessing lexical production and comprehension using standard tests. This study, conducted on a sample of 15 CWS and 15 CWNS with an average age of 35 months, reported a deficit in the development of expressive vocabulary in CWS. According to the authors, this result could reflect a strategy of avoiding the words that elicit more disfluencies and would not necessarily represent a difficulty in finding the correct lexical target by the CWS (see also Bloodstein, Ratner & Brundage, 2021). In this regard, a study by Quartarone, Bonichini, Natarelli, Pisciotta & Zmarich (2016) suggests that delays in lexical development may be present in children who subsequently exhibit stuttering well before the onset of stuttering, and children whose stuttering becomes persistent are significantly more delayed and exhibit more SLDs than those whose stuttering will recover spontaneously. Fourteen children, all born in a family where one or both relatives were PWS, were audio and video recorded at 24 months while performing an informal interview and a phonetic test. Adding to this, the MacArthur-Bates CDI was compiled by the parents. To determine the final clinical outcome of each child, a structured telephone interview was conducted with the parents after an average of 40 months from the stuttering onset (range: 20-68 months), revealing that 4 children had become persistent stutterers (CWS), and 10 had recovered spontaneously (CWNS). By comparing the lexical age of the subjects against age-referenced normative values when they were 24 months old, both groups of children presented a delay in lexical age. Still, future CWS were significantly more delayed ($M = -3.42$ months) than the future CWNS ($M = -1.90$ months), also producing a significantly higher number of SLD (3.01 % vs 0.75 %).

The studies by Anderson, Conture (2000) and by Ntourou *et al.* (2011), also reveal greater weaknesses of CWS than CWNS on all measures of receptive and expressive vocabulary. The differences between the two groups are probably attributable to the poor phonological working memory exhibited by the CWS (Gathercole, 2006; Gathercole *et al.*, 1999) and to the limitation of attention capacities, frequently found in these subjects. Attention, in particular, is considered an indispensable factor for semantic-lexical development, as it allows children to

grasp the correlation between each new word and its semantic referent (Spaulding, Plante & Vance, 2008; Finneran, Francis & Leonard, 2009).

Regarding the characteristics of the CWS group alone, the study by Coulter *et al.* (2009) highlights significant dissociations between the receptive and expressive vocabulary, demonstrating how the former is on average less developed than the latter in subjects with stuttering. This evidence, although in line with what previously reported by Anderson, Conture (2000), constitutes a unique and unexpected result, if we consider that, in the evolutionary stages of language development, verbal comprehension precedes and influences production. Interesting studies on the priming effect were then carried out to analyse the temporal course of linguistic processing, from the stage of perception of the stimulus to its articulation, through the retrieval of the lemma and the subsequent coding of the corresponding lexeme (Levelt, 1989; Bock, Levelt, 1994). A first experiment showed that CWNS usually name the target more quickly when preceded by a semantically related prime, contrary to CWS who do not seem to benefit from it and who are instead more facilitated by syntactic priming (Pellowski, Conture, 2005). This result seems to confirm the hypothesis put forward by Hall (2004) according to which important dissociations between syntactic and semantic-lexical skills in the child could affect the structuring of the utterance, interrupting or slowing down language planning and consequently leading to an increase in disfluent episodes.

Finally, the results of the aforementioned research and those of six other experimental studies were compared in the review by Luckman *et al.* (2020), which highlights that the performance of CWS between the ages of 2 and 8 is on average one standard deviation below the CWNS average value in expressive vocabulary, although it never falls into the deficit range. Furthermore, the lexical variety exhibited by CWS, measured in a conversational context, was found to be comparable to the group of CWNS. These data suggest that, during spontaneous interaction, CWS tend to select the words that best suit their coding skills, and instead find themselves in difficulty in standard tests, when they are asked to retrieve a specific lexical target.

1.4.5 Discourse level

The assessment of the child's narrative skills is of fundamental importance in the clinical setting as it investigates both the micro-processing aspects of expressive language and the macro-processing ones (Nippold, 2010; Nippold, 2012). Furthermore, narration can highlight more nuanced linguistic problems not directly identifiable by common standard tests (Roth, Spekman, 1986; Westby, 1979; Nippold, 2012).

In an initial study of the narrative abilities of school aged male CWS, Nippold, Schwarz & Jescheniak (1992) found no significant differences between them and CWNS peers in the dimensions of narrative length and complexity, of the structural integrity of the reported episodes and of the grammatical components of the story. Similar conclusions were reported by Scott, Healey and Norris (1995) on

school-age CWS who were given a retelling task, and more recently by Bajaj (2007), focused on the analysis of the narration ability on an iconic stimulus (cf. *Frog Story*).

An interesting study by Weiss, Zebrovsky (1994), conducted on 16 preschool CWS and CWNS, tested the narrative skills of the sample in the retelling task of 2 stories to listeners who already knew them or not, and in the telling of 3 invented stories. The study highlighted how, contrary to CWNS, CWS are not used to resorting to longer or richer utterances in reporting the story to those who do not know it, although they still manage to transmit the main basic information. This result suggests, on the one hand, the presence of a greater narrative effectiveness in CWS, and on the other reveals a certain pragmatic difficulty related to the inferential sphere and the amount of information to convey to each interlocutor.

1.5 Experimental hypotheses and aims

The present study aims at investigating possible correlations among stuttering severity, CA, and linguistic skills, in a group of CWS. First of all, it is assumed that CWS could develop a negative CA not only because of fluency problems, but as a result of other speech and language difficulties different from stuttering they might have, although not necessarily of clinical relevance, like the difficulties evidenced by scoring low at language batteries. Second, since we expect that negative CA increases together with the number of stressful events associated with stuttering, we expect that it could increase along with severity, age at the time of the recording, age at stuttering onset, and the amount of time elapsed from the stuttering onset.

2. Method

The study involved 18 children (15 males, 3 females) aged between 4 years, zero months, and 6 years, 11 months (mean age 5 years, 5 months), diagnosed as stutterers by certified speech therapists, but not yet treated. The onset of stuttering, as reported by their parents, happened at a mean age of 43 months (SD 14.3). They were Italian monolinguals, living in Veneto, without other certified concomitant disorders. The *SSI-4* (Riley, 2009) was used to assess stuttering severity, the *BVL 4-12* (a psycholinguistic battery with wide coverage, Marini *et al.*, 2015), for language and speech skills assessment in production and perception starting from four years of age, and the *KiddyCAT* (Vanryckeghem & Brutten, 2022, which is the Italian adaptation of Vanryckeghem & Brutten, 2007), for the evaluation of the CA from 3 to 6 years of age. In addition to signing the consent to participation and to data treatment, parents completed a questionnaire about the subject's medical history and family information.

The tests were administered to each child through two meetings lasting about 2 hours each, at children's home or at the speech therapist's office. All sessions were audio- and video-recorded with professional instruments. The video recordings were carried out using the Sony HDR-CX115E TV camera, while audio recordings

were acquired using the the Edirol-R09 HR, placed approximately 20-30 cm away from the child's mouth, sampling the acoustic signal at 44kHz and 16 bits.

We used the normative samples contained in the KiddyCAT (Vanryckeghem *et al.*, 2022) and in the BVL to compare the experimental subjects with a valid sample of CWNS. For this reason, the scores are expressed as z-scores.

As to severity level, there were 3 severe, 9 moderate, 4 mild and 2 very mild subjects.

3. Results

The average score of subjects on the KiddyCAT is 2.82 (DS=2.18; CWS' mean in Vanryckeghem, Brutten, 2022: 4.37; SD: 2.56) and it is not significantly different from the mean score of CWNS (0.72; SD: 1.19). Only 4 children out of 18 have obtained a score that is indicative of a negative CA.

Regarding speech and language skills, all cautions were taken in order to exclude fluency difficulties from the calculation of the BVL scores. For instance, in the Naming and in the Sentence Completion tasks, children were given a wait time of up to 10 seconds before responding (unlike children with normal fluency who are given 10 seconds in total). The performance was considered valid if the subject, despite the disfluencies, completed the response within 10 seconds from starting. In the Narrative Fluency task and in the Multilevel Assessment of Narrative Speech task, we excluded Stuttering-like Disfluencies (SLD), such as syllable repetitions, in the count of units produced. In narrative fluency and in all other tests with time limits (Semantic Fluency and Phonological Fluency), we tried to be flexible with the imposed limit, granting the child additional time in case he lost time due to SLDs.

As to the results at the BVL battery, a detailed exposition of the individual performances at each sub-test is beyond the scope of the present paper. We summarise the results in table 1, where, for each sub-test, the number of children who obtained a performance to be classified in five performance ranges was calculated. These are: below -2 SD (deficit level), between -2 and -1 SD (lower than normal level), between -1 and +1 SD (level corresponding to the norm), between +1 and +2 SD (above the normal level) and above +2SD (far above the norm level).

Considering all the tests of the BVL battery, most of the participants (around 70 %) have obtained scores that fall within the average expected for their age (between -1 SD and +1 SD). The second following level is "lower than normal" (13.09 %), followed by "upper than normal" (8.20 %), then deficit level (7.34 %), and lastly, "far above the norm level" (1.27 %). Summing up the two levels under the norm level, we obtain around 20 % of all the children, while summing up the two levels above the norm, we obtain around 10 % of all the children.

The individual tests where most of the children scored less than -1 DS were (in percentages out of the total sample): Repetition of sentences (43.8 %); Repetition of words (44.4 %); Grammatical judgement (38.9 %); Articulation (naming) (33.3 %,

with 4 children scoring below -2 SD and 2 children scoring between -1 SD and -2 SD); Non-words repetition (33.3 %, with 3 children scoring below -2 SD and 3 children scoring between -1 SD and -2 SD).

Table 1 - *No. of children in each of the five performance levels considered for each test of BVL 4-12*

	< -2	-2≤DS<-1	-1≤DS≤1	1<DS≤2	> 2
<i>PRODUCTION</i>					
Articulation	4	2	11	1	0
Naming	0	1	13	4	0
Semantic Fluency	0	1	12	3	2
Phonologic Fluency	2	3	9	2	0
Sentence Completion	1	1	14	1	0
No. of produced Words	0	2	14	1	1
Narrative Fluency	1	5	10	2	0
MLU	2	1	13	2	0
<i>COMPREHENSION</i>					
Phonologic Discrimination	0	1	17	0	0
Lexical Comprehension	0	2	13	1	0
Grammatical comprehension	0	1	15	2	0
Grammatical Judgement	2	5	10	1	0
Idiomatic Expressions	1	3	10	4	0
Linguistic Prosody	1	1	14	1	1
Affective Prosody	0	0	18	0	0
<i>REPETITION</i>					
Word Repetition	2	6	10	0	0
Non-Word Repetition	3	3	12	0	0
Sentence Repetition	4	3	8	1	0
ALL THE TASKS	23 (7.34 %)	41 (13.09 %)	223 (70.34 %)	26 (8.20 %)	4 (1.27 %)

Looking at the results of the three repetition tasks, the only one subject performing above mean level is outnumbered by the many subjects performing below the mean level, with substantial percentages scoring below two SDs, which is considered of clinical interest. Statistical investigations of correlation between the various performances at the individual test of the BVL 4-12 were conducted. The correlation index *Rho* for Spearman's ranks was used, a non-parametric statistical measure of correlation suitable for small samples, which do not follow a normal distribution, as in the case of the present study. The results of the analyses highlighted:

- A positive correlation between Semantic Fluency and Narrative Fluency ($Rho=0.57$, $p<0.05$);
- Positive correlations between Non-Word Repetition and: Grammatical Judgment ($Rho=0.55$, $p<0.05$), Articulation ($Rho=0.60$, $p<0.01$); Phonological discrimination ($Rho=0.69$, $p<0.01$), total mean of the production tests ($Rho=0.63$, $p<0.01$), total mean of all the other linguistic tests of the battery ($Rho=0.78$, $p<0.001$);
- Positive correlations between the Phonological Discrimination and: Non-Word Repetition ($Rho=0.69$, $p<0.01$), Narrative Fluency ($Rho=0.69$, $p<0.05$), Articulation ($Rho=0.54$, $p<0.05$), total mean of the production tests ($Rho=0.67$, $p<0.01$), total mean of all the other linguistic tests of the battery ($Rho=0.64$, $p<0.01$);
- A positive correlation between the total mean of the production tests and the total mean of the comprehension tests ($Rho=0.74$, $p<0.001$).

No significant association was found between the individual scores, expressed in zeta-scores and averaged from all BVL 4-12 tests, and the individual raw stuttering severity scores from SSI-4 ($Rho=-0.07$, $p=0.722$), nor between the individual scores, expressed in zeta-scores and averaged from all BVL 4-12 tests, and the individual scores obtained from the KiddyCAT ($Rho=-0.07$, $p=0.79$). We also tested the degree of correlation between the individual raw stuttering severity scores from SSI-4 and the individual zeta-scores on every single test of the BVL 4-12. Although most of the correlations did not show statistical significance, there were two notable exceptions. For the BVL 4-12 Naming test, a remarkably significant and strong negative correlation emerged ($Rho=-0.571$, $p=0.005$). This indicates that as scores on the naming test increase, stuttering severity tend to decrease monotonically. Second, the Semantic Fluency test showed a negative correlation with SSI-4 scores ($Rho=-0.481$, $p=0.023$), suggesting that an increase in Semantic Fluency scores is associated with a decrease in stuttering severity.

Stuttering severity, as measured by both the individual raw scores of SSI-4 and by individual scores derived from the frequency subscale of dysfluencies, also did not correlate significantly with individual CA scores.

Finally, there were no statistically significant correlations between KiddyCAT scores and a) chronological age, b) age at stuttering onset, c) number of months elapsed since stuttering onset.

4. Discussion and conclusions

Our results turn out to be in line with the current literature, showing a substantial similarity between the speech and language skills of stuttering and nonstuttering peers, except for the very bad performances (<-2 SD) in the articulation task and in the word and non-word repetition tasks. The present study did not find any significant correlation between CA and stuttering severity (as in Winter, Byrd, 2021), CA and speech and language skills, CA and time of onset, or CA and time

elapsed from the stuttering onset to the experimental session. In our opinion, the absence of correlations between CA and stuttering severity could be explained by the unusual positive communicative attitude held by most of the subjects, with a mean score below the mean for CWS in Vanryckeghem *et al.* (2022), and by the absence of any certified comorbidity with other speech and language disorders. Here we provisionally advance an hypothesis inspired by the results of Choi, Hurd & Kim (2022) on the level of awareness of disfluent speech in CWNS, that positive CA may be a consequence of not having a speech-language monitoring system sufficiently developed, causing most of CWS not to be aware of their problems and consequently not to be negatively affected in their CA.

The unexpected result that the most severe CWS obtained the lowest scores on the BVL 4-12 Naming subtest and the Semantic Fluency subtest seems to suggest for these subjects a possible problem in the lexical access process, but until it is possible to exclude a possible methodological bias related to the difficulty of the subjects more disfluent in respecting the time limits of these tasks (and to the related difficulty for the present authors to compensate for this), we prefer to suspend any further consideration.

Our final consideration involves the subjects' poor performance in repetition tasks, particularly NWR, regardless of severity scores. This result is in line with other studies (Pelczarski, Yaruss, 2016; Sasisekaran, Weathers, 2019) and it is consistent with the evidence of the high comorbidity of stuttering with Speech Sound Disorders (SSD). Interestingly, Gerwin *et al.* (2022) found that CWS with SSD perform significantly lower than CWS without SSD in a task of non-word repetition (NWR). NWR has also been claimed by Graf-Estes *et al.* (2007) and Coady, Evans (2008) as a powerful index for SLI. Regrettably, bad performance on NWR could be due to different causes (Krishnan *et al.*, 2017), because NWR reflects a variety of speech and language skills, including lexical access (e.g. Rubenstein *et al.* 1970), motor planning (e.g. Yoss, Darley 1974), phonological processing (e.g. Snowling 1981), phonological memory (e.g. Gathercole, Baddeley, 1989; Bowers, Bowers, Hudock & Ramsdell-Hudock, 2018), and the task also intercepts the processes of linguistic perception, phonological assembly and articulatory behaviour (see the review of Anderson *et al.*, 2019 pointing to CWS' weakness in executive functions; see also Sasisekaran, Basu, 2017). However, new light has been shed on NWR by Krishnan *et al.* (2017), claiming this task in children to be the reflection of the ability to compute and perform the sensorimotor conversions necessary to produce new words. This explanation matches the theories which interpret stuttering as a consequence of sensory-motor integration deficits (Speech Motor Control models based on efferent copy or hybrid feedback: Max *et al.*, 2004; Hickock *et al.*, 2011; Tian, Poeppel, 2012; Civier *et al.*, 2013; for a review, see Bradshaw *et al.*, 2021). In fact, Bowers *et al.* (2018: 94) state that "cognitive-behavioral differences in NWR and dual-tasks are consistent with the involvement of neurocognitive networks related to executive function and sensorimotor integration in phonological working memory", and "mistimed or 'noisy' dorsal stream sensorimotor integration

previously proposed to cause stutter-typical disfluencies (i.e., part-word repetitions, prolongations, and blocks) may also account for load dependent differences on nonword repetition tasks in CWS and AWS". Last, but not least, these results could have a precious clinical value: Walsh *et al.* (2021) argued that prognostic index of chronic stuttering are: familiarity, low scores on standardized phonetic-phonological assessment tests, high frequency of SLD in spontaneous speech, and *low scores on NWR* (italics are of the present authors), and a prognostic value is explicitly assigned to NWR by Spencer, Weber-Fox (2014), Leech *et al.* (2017) and Mohan, Weber (2015).

We conclude by admitting that a limitation of the present research is the absence of a control group made up of CWNS matched by age and SES. This limit is to be understood as a suggestion for a better continuation of the project. Another proposal for the future is to increasingly expand the group of participants and diversify it, also recruiting stuttering subjects with certified comorbidity with language and/or speech disorders. A further extension of this study could concern ascertaining the status of persistent stuttering years after the performances of the subject of this contribution, and then using a statistical regression model to verify how the clinical outcome can predict the scores of linguistic proficiency and CA that we have presented here. This would help to clarify some results which are currently difficult to interpret, because they are potentially the result of two different populations: those of stutterers who will recover and those who will persist. That the two populations are distinguishable on the basis of behavioral data is a fact now shared by the majority of scholars (see Bloodstein, Bernstein-Ratner & Brundage, 2021; Yairi, Seery, 2023), and much of the current research on stuttering is aimed at identifying children predisposed to persistence early on the basis of some behavioral measures, which can then be used in a clinical setting. In fact, stuttering is characterized by having a very early average age of onset (33 months), but also a very high spontaneous recovery rate (around 90 %), which occurs, in 95 % of children who had begun to stutter, within the first 4 years of onset (Yairi, Ambrose, 2005). In light of these data, most of the CWS examined in this study could therefore currently be recovered, since when they were examined they had already been stuttering for many months (especially the older ones) and many more months have passed from the day of recording to date.

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