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TITOLO DELLA TESI: Communicative-pragmatic abilities in atypical development: Hearing Loss and Autism Spectrum Disorder

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*“There are no telepaths in this universe,
I think, but there are empathics,
with languages so silent that they may as well be sharing thoughts.”*

Alle mie donne, piccole e grandi.

Summary

| | |
|--|-----------|
| 1. General introduction | 9 |
| 1.1. Aims of the thesis | 9 |
| 1.2. Theoretical frameworks on communication | 13 |
| 1.2.1. Speech acts theory | 13 |
| 1.2.2. Pragmatic inferential model | 15 |
| 1.2.3. Cognitive pragmatic theory | 17 |
| 1.2.4. Narrative ability..... | 20 |
| PART A | 22 |
| Acquired physiological deficit: Hearing loss | 22 |
| 2. Introduction | 24 |
| 2.1. Communicative-pragmatic ability in children with cochlear implant | 26 |
| 3. Study one: Pragmatic language ability in children with early cochlear implants | 33 |
| 3.1. Aims and hypothesis | 33 |
| 3.2. Material and methods..... | 34 |
| 3.2.1. Participants | 34 |
| 3.2.2. Material | 36 |
| 3.2.3. Procedure..... | 39 |

| | |
|---|-----------|
| 3.2.4. Data analyses..... | 39 |
| 3.3. Results..... | 40 |
| 3.3.1. Pragmatic ability assessment..... | 40 |
| 3.3.2. Role of age, gender, non-verbal intelligence, and age at implantation at implantation | 45 |
| 3.4. Discussion..... | 45 |
| 4. Study two: Multimodal pragmatic abilities in children with early cochlear implants..... | 52 |
| 4.1. Aims and hypothesis | 52 |
| 4.2. Methods | 53 |
| 4.2.1 Participants..... | 53 |
| 4.2.2. Material | 55 |
| 4.2.3. Procedure..... | 59 |
| 4.2.4. Data Analysis | 59 |
| 4.3. Results..... | 61 |
| 4.3.1. Multimodal pragmatic ability..... | 61 |
| 4.3.2. Role of implantation and non-verbal intelligence | 65 |
| 4.4. Discussion..... | 65 |
| PART B | 73 |
| Neurodevelopmental disorders: Autism spectrum disorder..... | 73 |
| 5. Introduction | 75 |

| | |
|---|------------|
| 5.1. Communication in adolescents with autism spectrum disorder..... | 78 |
| 6. Study three: Narrative ability in adolescents with autism spectrum disorder receiving the Cognitive-Pragmatic Treatment | 84 |
| 6.1. Aims and hypothesis | 84 |
| 6.2. Material and Methods | 86 |
| 6.2.1. Participants | 86 |
| 6.2.2. The rehabilitative program: CPT for adolescents | 87 |
| 6.2.3. Assessment measures | 91 |
| 6.2.4. Data analysis | 97 |
| 6.3. Results..... | 97 |
| 6.3.1. Principal component analysis..... | 97 |
| 6.3.2. Narrative assessment | 99 |
| 6.3.3. Cognitive skills assessment..... | 102 |
| 6.3.4. Theory of Mind assessment..... | 102 |
| 6.4. Discussion..... | 103 |
| 7. General discussion | 110 |
| Appendix A: ABaCo's Item examples..... | 115 |
| References | 120 |

1. General introduction

1.1. Aims of the thesis

The present doctoral thesis explores pragmatic and narrative aspects of communication in atypical development, specifically in disorders caused by physiological factors (e.g., hearing loss) and neurodevelopmental disorders (e.g., autism spectrum disorder). To function properly, communicative ability relies on: *syntax* - the grammatical structure of a sentence; *semantics* - the meaning of words and sentences; and *pragmatics* - the use of language in a specific context and the speaker's intention (Levinson, 1983; Morris, 1938). Pragmatics is a multimodal ability; it refers to the capacity to appropriately use language and other expressive means (e.g., gestures, facial expressions, tone of voice, proxemics) to convey meaning in a specific context (Bara, 2010; Levinson & Holler, 2014). Communicative-pragmatic ability comprises: the ability to manage a conversation by maintaining a discourse topic, respecting turn-taking, and complying with social and conversational norms (e.g., Grice's Maxims; Grice, 1975, 1989); the ability to use language appropriate for the interlocutors (e.g., age and social status); and the ability to go beyond the literal meaning of an utterance (e.g., metaphors, proverbs, irony; Bates, 1976; Cummings, 2005; Grice, 1989).

Narrative competence is also part of communicative ability. It can be described as the capacity to present events that are connected causally and temporally in a comprehensible way for the listener, while considering shared knowledge and additional information that might be needed for complete comprehension (Capps et al., 2000). To produce a good story and discourse, the speaker must organize and structure the narrative in a cohesive and coherent manner by exploiting linguistic devices that will make the account easy to understand in all its parts and contents (Rumpf et al., 2012).

There is abundant literature on how communicative ability evolves in typical development (Airenti, 2017; Cekaite, 2012; Matthews et al., 2018), but less is known about this ability in atypical development.

Communicative ability can be impaired when:

- there is an acquired physiological state: a deficit in physiological function that does not allow typical development;
- there is a neurodevelopmental disorder: a disorder or disease from birth or that manifests later, causing communicative impairment, which is a symptom of the disorder.

For this thesis, I have selected a case for each factor that can cause communicative impairment. I focus on hearing loss in relation to acquired physiological state. A hearing deficit can hinder the perception of external sounds and slow language acquisition and development (Paatsch et al., 2017). For the other factor, I examine autism spectrum disorder (ASD) whose main symptoms include communicative impairment (APA, 2013).

Communicative impairment can be improved by treatment following accurate assessment and comprehension of the complexity of the deficit. Individuals with residual hearing, for instance, are usually fitted with a hearing aid, while those with profound hearing deficit may receive a cochlear implant (CI). A cochlear implant is an electronic device inserted near the cochlea that transforms external sounds into electrical stimuli for transmission to the auditory nerve (Clark, 2015). The device has proven effective in promoting language acquisition and communicative-pragmatic ability (Crowe & Dammeyer, 2021), especially in children who received an implant at a very early age (Yoshinaga-Itano et al., 2020). Individuals with hearing loss usually undergo speech therapy (e.g., auditory-verbal therapy) to develop their communicative-pragmatic ability (Binos et

al., 2021). Somewhat similarly, individuals diagnosed with ASD receive therapy to help them perform key activities of everyday life autonomously (e.g., dressing and self-hygiene) or improve their basic linguistic skills (Wickstrom et al., 2021). Those with sufficient linguistic abilities may be eligible for treatment focused more on gaining social (Choque Olsson et al., 2017; Dekker et al., 2019; Matthews et al., 2020) and pragmatic skills (Gabbatore et al., 2021).

However, the literature on the communicative ability, specifically pragmatics and narrative, of these clinical populations is scarce and inconclusive. Assessment of communicative-pragmatic ability in children who received a cochlear implant early in life (<24 months) has not taken into consideration all expressive means (linguistic, extralinguistic, paralinguistic). Furthermore, debate surrounds the role of age at implantation. *Part A* of the present thesis regards the assessment of pragmatic language in children with hearing loss and early cochlear implantation, with specific attention to the role of age at first implantation (*Part A: Study one*). This is followed by an analysis of multimodal pragmatics in children with early cochlear implantation and the effect of age at implantation on pragmatics (*Part A: Study two*).

Similarly, the majority of the existing studies on the assessment of communicative impairment and treatment of communicative-pragmatic ability in ASD focus on school-aged children. Very few studies have investigated rehabilitative treatment for later stages of development, i.e., adolescence (Parsons et al., 2017). Gabbatore and colleagues (2021) studied the effect of pragmatic treatment on pragmatic skills of a group of adolescents with ASD and McCabe et al. (2017) found improvement in narrative production in adolescents and young adults with ASD after receiving a parental-mediated intervention. *Part B (Study three)* of the dissertation is dedicated to an assessment of narrative abilities before and after rehabilitative treatment in adolescents diagnosed with ASD (*Part B: Study three*).

The present dissertation starts with an overview of the main theoretical frameworks for communicative ability. I refer to the inferential model and within this general framework specifically to cognitive pragmatic theory (Airenti et al., 1993a, 1993b; Bara, 2010), which I will adopt as the theoretical background in *Study two* and *Study three*.

In *Part A: Study one* entitled “Children with hearing impairment and early cochlear implant: a pragmatic assessment” by Hilviu, Parola, Vivaldo, Di Lisi, Consolino, and Bosco (2021), we focused on language, which is conventionally investigated within the context of pragmatics. We investigated the development of pragmatic language ability in school-aged children with severe-to-profound hearing loss fitted early with a cochlear implant (by age 24 months). Many studies on pragmatic language in this clinical population have produced inconsistent results. The aim of the present study was to determine how this ability develops in children with a cochlear implant for hearing impairment and the effect of early implantation on performance.

Part A: Study two, entitled “Development of communicative-pragmatic abilities in children with early cochlear implant” by Parola, Hilviu, Vivaldo, Di Lisi, Consolino, and Bosco (under review), investigated multimodal pragmatics through a broad assessment of communicative-pragmatic ability in children with a cochlear implant for hearing impairment and the effect of the implant on their global communicative performance. Previous studies have focused on the linguistic and the conversational aspects of pragmatics, frequently neglecting the fact that communication is carried also by expressive means: extralinguistic, such as gestures, and paralinguistic, such as tone of voice and cues (Bara, 2010).

Part B comprises *Study three* “Can narrative skills improve in autism spectrum disorder? A preliminary study with verbally fluent adolescents receiving the Cognitive Pragmatic Treatment” by Hilviu, Frau, Bosco, Marini, and Gabbatore (under review). The study is an analysis of the assessment of narrative abilities in adolescents with ASD before

and after administration of cognitive-pragmatic treatment (CPT) adapted for adolescents, a rehabilitation program specifically designed to treat communicative-pragmatic and social and cognitive abilities (Gabbatore et al., 2021). With this study we wanted to assess narrative skills and evaluate the effect of CPT on narrative ability.

1.2. Theoretical frameworks on communication

1.2.1. Speech acts theory

Wittgenstein (1953) was the first scholar to interpret language and communication differently, namely as a game regulated by internal rules shared by interlocutors. In the same years, Austin (1962) noted the existence of *performative utterances*, i.e., utterances that, when proffered in specific contexts and circumstances, behave like actions. He then proposed a *Theory of Speech Acts* where he identified three components that jointly appear when communicating: the *locutionary act* concerns the expression of an utterance, the *illocutionary act* concerns the intention of the speaker and finally, the *perlocutionary act* concerns the effects that the speaker wants to induce in the listener. The theory of Austin was later reframed by Searle (1975) who proposed a *Taxonomy of the Illocutionary Acts* in order to categorize different speech acts. Building on these initial contributions, the Shannon–Weaver model of communication, in which communication is defined as a transfer of information¹, is now considered an incomplete way to describe human communication. The aforementioned authors helped to create the concept that the meaning of an interaction is not given but rather the result of a cooperative interaction between two interlocutors in a

¹ The *Shannon–Weaver model of communication* describes communication as the transfer of information where the message containing the information is first coded by the sender and then decoded by the recipient (Shannon & Weaver, 1949).

specific context; accordingly, communication can be seen as a kind of action. The activity was described in more detail by Grice in his *Cooperative Principle* (1975):

“*Make your contribution such as is required, at the stage at which it occurs, by the accepted purpose or direction of the talk exchange in which you are engaged.*” (cit. Grice, 1975, p. 45).

This principle is regulated by Grice’s four maxims (Grice, 1975, 1989) specifying that the contribution provided by the speaker should be adequately informative (*quantity*), truthful (*quality*), relevant to the topic (*relation*), brief and not ambiguous (*manner*). These maxims allow interlocutors to make inferences and understand the meaning of what is proffered. He suggested that every communicative act has a literal meaning and an intended meaning, which must be inferred and that depends on the speaker’s intentions (Grice, 1989). The gap between what is said and what is intended is filled by individuals via specific processes called *implicatures*. For instance, in [1]:

[1] A: *Do you like cats?*

B: *I have four of them!*

B is communicating more than only the literal meaning. Specifically, B is saying that she/he loves cats and that she/he has not just one but four of them. The theory of communication proposed by Grice grounds its definition on the mutual understanding that takes place when interlocutors adhere to the principle of cooperation that guides the inferential processes necessary to fill the gap between the literal meaning and the intended meaning of an utterance.

1.2.2. Pragmatic inferential model

Some scholars might argue that the aforementioned authors (i.e., Austin, Grice, etc.) did not interpret their theories in terms of psychological and cognitive processes of language, which were rather taken into consideration by other authors (Sbisà, 2018).

The *principle of relevance* was advanced by Sperber and Wilson (1986) within the Relevance theory and argues that individuals address their attention to an utterance based on its relevance to context. Sperber and Wilson state that a certain quantity of information is already possessed by individuals thanks to their past experiences while other information is available in the context (1986). Relevance is defined as the ability to recognize in the context at hand stimuli/information that can revise beliefs already held as true and produce new information (new beliefs). This ability is limited, however, by the cognitive effort needed to process the given information, so individuals select where to direct their attention and effort (Bambini, 2017). Despite its simplicity, Relevance theory seems to be excessively based on context and unable to completely explain how the stability of meaning is produced in order for interlocutors to understand what is proffered (Anolli, 2006).

The ability to fill a gap using inferences in order to go beyond the literal meaning and understand the intended meaning also characterizes the metaphorical language. Metaphor is a figure of speech and is the description of an entity in terms of another entity (Bambini, 2017; Ervas & Gola, 2016).

[2] *Tom is a tiger!*

In example [2], Tom is described as a tiger and likened with its characteristic feature, determination. Metaphors use a vehicle (the term used metaphorically) to describe a topic (the object of the description). In [2], *Tom* is the topic and *tiger* is the vehicle. Therefore, metaphor, as a figure of speech, is both the vehicle and topic in their interconnection, with

possible emergent properties that do not belong neither to the vehicle nor to the topic semantic domains (Wilson & Carston, 2006, 2008). Metaphors are based on the fact that topic and vehicle share some features and are on the same ground. This allows individuals to use some features of the vehicle to describe the topic.

According to Grice (1975), metaphors represent cases of overt violations of the *maxim of quality*, thus causing in this way an implicature (Domaneschi & Bambini, 2020). From a cognitive point of view, metaphors are conceptual processes: they are defined by a mapping between two concepts, a *source domain*, which is usually the one more familiar and concrete and the *target domain*, which tends to be more abstract and intangible (Lakoff & Johnson, 1980)². In this way, metaphors can also support the acquisition of new abstract concepts. In relevance theory, metaphors are conceptual processes where concepts undergo an adjustment (broadening or narrowing the linguistically encoded concept) in order to construct an *ad hoc* concept (Wilson & Carston, 2006). How exactly metaphors are processed is still debated (Bambini, Bertini, et al., 2016; Noveck, 2018): some authors claim that it takes more time to understand a metaphor than literal communication (indirect access) (Grice, 1975; Searle, 1979). The reason why is the longer and more complex inferential chain typical of non-literal language (Janus & Bever, 1985). Other authors state that in some circumstances the context helps individuals to understand non-literal language as if it were literal in terms of time (direct access) (Gibbs, 1992, 1994). Recent empirical data seem to support the indirect access hypothesis (Weiland et al., 2014). Comprehension of metaphors requires finely developed complex linguistic and cognitive skills as prerequisite. Given the greater complexity of metaphors compared to literal language (Bambini et al., 2013; Bambini, Bott, et al., 2021; Bosco, Vallana, et al., 2012; Noveck et al., 2001), they tend to

² Nevertheless, many critics might point out that the relation between source (concrete) and target (abstract) domain is not always unidirectional but it can be also bidirectional: concrete-to-abstract, abstract to-concrete, and concrete-to-concrete, such as in the famous metaphor “Juliet is the sun” both the source and the target are familiar and concrete (Freeman, 2017; Goodblatt & Glicksohn, 2017).

be fully understood only in late childhood (Winner et al., 1976). Nevertheless, some recent studies suggest that at age 3-4 years, children start to understand some metaphors (Di Paola et al., 2020). However, the comprehension of metaphors seems to be affected by numerous other factors (Bambini et al., 2011; Kintsch & Bowles, 2002) such as the level of familiarity, namely, the frequency of occurrence of the figurative expression that the child possesses with the conceptual domain used by the metaphor (Rataj, 2014; Vosniadou & Ortony, 1983), the aptness, which is the extent of features of the topic that the vehicle can capture (Blasko & Connine, 1993; Damerall & Kellogg, 2016), and by the differences in how metaphors are presented (embedded in a story or isolated) (Özçalışkan, 2008).

1.2.3. Cognitive pragmatic theory

Within the pragmatic inferential model, the *cognitive-pragmatic theory* (Airenti et al., 1993a, 1993b; Bara, 2010) focuses on the cognitive and inferential processes underlying communication. In the present dissertation, *Study two* and *Study three* are based on this theoretical framework. According to cognitive-pragmatic theory, communication is the ability to use linguistic and extralinguistic means, such as gestures in combination with paralinguistic cues (e.g., tone of voice, intonation, etc.) to convey meaning in a given context (Bara, 2010). Individuals use language, which is a complex system of symbols (e.g., words) where the latter can be combined to create new and infinite meanings (Bara, 2010) to communicate. Another expressive means is extralinguistic ability (e.g., gestures), which is based on a set of symbols that cannot be divided into subcomponents and cannot be combined to create new meanings. Previous studies have indicated that linguistic and extralinguistic abilities follow the same path of emergence in children (Bara et al., 2000) and that these expressive modalities are understood in an equivalent way (Bucciarelli et al., 2003). Finally, utterances and gestures can be accompanied by paralinguistic cues (e.g.,

prosody, facial expressions, proxemics, etc.), which can emphasize or change the meaning of a speaker's words. Cognitive pragmatic theory is based on the assumption that the literal meaning of a sentence is insufficient for understanding it completely. The speaker's intended meaning needs to be reconstructed to fully understand what the speaker wanted to communicate. In brief, communication requires cooperation between at least two conversational partners. Cooperation is possible only when:

- the interlocutors share knowledge of the *behavioural game*, which regulates the actions in an interaction;
- the interlocutors adhere to the rules of the *conversational game*, namely, the rules governing comprehension and generation communicative acts.

[3] A: *Do you have 30 euros?*

B: *I forgot my wallet at home!*

In example [3]³, B understood that A wanted to know not only if B had 30 euros but also if B was willing to lend the sum of money to A. In this exchange, B understood and related the communicative act expressed by A to a specific behavioural game, i.e., *lending money*. For B to understand what A is trying to communicate through different expressive means, B must be able to relate this expression to the game in which it is played.

Moreover, when producing and understanding a communicative act, the interlocutors go through a series of phases starting from the comprehension of the literal meaning to the comprehension of the intended meaning: communication is not an “all or nothing” activity. The *conversational game* regulates the production and understanding of a communicative

³ This example was taken from the handbook of the Assessment Battery of Communication (Angeleri et al., 2015)

act. It consists of a set of rules that define the inferential processes involved in the understanding and the production of the act.

[4] A: *Good job!*

In example [4], B can understand the literal meaning of what A has proffered without having understood the intention underlying the communicative act. However, conversational rules can also be violated by producing *non-standard communicative acts*. While in *standard communicative acts* what the speaker communicates is coherent with his/her beliefs and mental states, in non-standard acts what is expressed is not coherent with the mental states of the speaker and with the beliefs shared by both interlocutors. For example, irony, deception, and figurative language (e.g., metaphors) are instances of non-standard communication. In such cases, the inferential chain to understand the non-standard communicative act is longer than the one necessary for literal language, which adds difficulty to understanding the non-standard communicative act. (Bosco et al., 2013; Bosco et al., 2012; Bosco & Gabbatore, 2017a). Empirical findings suggest a differential ability (a trend of difficulty) of children (e.g., Bosco & Gabbatore, 2017a; Bosco & Bucciarelli, 2008) and subjects in clinical samples (e.g., Angeleri et al., 2008) to produce and understand communicative acts from the simplest to the most difficult: sincere, deceitful, and ironic acts.

Cognitive pragmatic theory is the theoretical background for a validated clinical test battery for the assessment of communicative abilities (ABaCo; Angeleri et al., 2012; Bosco et al., 2012; Sacco et al., 2008) that was adopted in the present dissertation (*Study two*), and a cognitively oriented pragmatic treatment (Cognitive Pragmatic Treatment; Gabbatore et al., 2015), which was used in *Study three* for the improvement of pragmatic abilities.

1.2.4. Narrative ability

Communication also includes the ability to produce narratives. Narrative ability is the ability to describe real life or fictive-based accounts of temporally and causally related events (Boudreau, 2008). In narratives, we refer not only to the “here and now” but also to the “there and then”, abstract concepts, imaginary things, and past or future events (Johnston, 2008). This ability is characterized by two levels of processing: micro- and macrolinguistic (Kintsch & van Dijk, 1978; Marini & Carlomagno, 2004). Microlinguistic processes allow for generation of the narrative at the lexical and the sentence level (i.e., intra-sentence processing). Macrolinguistic processes ensure pragmatic functionality of the discourse (i.e., between-sentence processing) through the use of cohesion devices and the generation of coherent episodes within the discourse (Marini & Carlomagno, 2004). Pragmatics is ensured by two narrative features: cohesion and coherence (Bambini, 2017; De Beaugrande & Dressler, 1981; Marini, 2008). Cohesion is defined as the connection between contiguous utterances using grammatical or lexical devices.

[5] “He got hurt but he is crying.”

In example [5], the right word to be used is “and” that provides the right derivation of a generalized conversational implicature (with a causal interpretation), rather than the conventional implicature possibly generated by the conjunction “but” (Grice, 1989). Coherence is defined by the correct conceptual organization of narrative production. In a coherent narrative, concepts are accessible and structural continuity is guaranteed (Bambini, 2017; Hickmann, 2003, 2004; Marini, 2008). Although conceptually different, cohesion and coherence are usually related to one another and their correct use produces a well-organized, comprehensible narrative (Bambini, 2017; Halliday & Hasan, 1976; Hickmann, 2004).

Narrative ability develops throughout life (Westerveld & Moran, 2013); its development appears to be related to multimodal (i.e., gesture, prosody, etc.) imitation in

young children at around age 3 to 4 years (Castillo et al., 2021; Vilá-Giménez et al., 2019). Studies investigating linguistic ability in narratives report that some microlinguistic components of grammatical processing (i.e., omission of morphosyntactic information and complete sentences) and macrolinguistic aspects (i.e., cohesion and coherence) change with advancing age (Marini, 2014). Unlike older children, young children (6-7 years old) produce utterances containing more omissions of morphosyntactic information and less syntactic completeness (Justice et al., 2006). At the macrolinguistic level, younger children produce more errors concerning referents (e.g., missing or misuses) (Loukusa et al., 2007). However, children already at age 5 can produce a coherent story (Marini, 2014). Finally, story informativeness increases with age (To et al., 2010); children of 6 to 8 years produce information-poor contents compared to slightly older children (8 to 10 years) (Marini, 2014). Narrative structure and contents continue to develop through late childhood to adolescence and adulthood (Berman & Slobin, 1994).

PART A

Acquired physiological deficit:

Hearing loss

2. Introduction

The World Health Organization (WHO) ranks hearing loss as the fourth cause of physiological disability in the world, with an approximate cost of US \$980 billion each year (WHO, 2021). It is estimated that in 2021, 432 million people suffer from some form of hearing loss (moderate or more severe) that impacts their quality of life (WHO, 2021). With the increase in global population and life expectancy, this number is expected to rise to 2.5 billion by 2050⁴ (WHO, 2021). While common causes of hearing loss (e.g., congenital rubella infection, and meningitis) are on the decline in some parts of the world, these and other infections are still widespread (e.g., otitis media, congenital cytomegalovirus and Ebola infection). Additional causes are ototoxicity and occupational or recreational noise exposure (WHO, 2021).

Hearing loss is measured and quantified by pure tone audiometry, the main hearing test conducted to determine type, degree (severity), and sidedness of hearing impairment and residual hearing ability. Hearing loss is categorized as (WHO, 2021):

- *Moderate* (35 to < 50 dB): some difficulty in taking part in a conversation;
- *Moderately severe* (50 to < 65 dB): marked difficulty in hearing speech and participating in a conversation;
- *Severe* (65 to < 80 dB): extremely difficult to hear conversational speech even with raised voices;
- *Profound* (80 to < 95 dB): inability to hear conversation;
- *Complete or total* (95 dB or greater): impossibility to hear speech and other external sounds.

⁴ These projections are based on evaluations that took into consideration global demographic changes.

Hearing loss can profoundly limit language acquisition and the development of other cognitive abilities, limiting social interactions and everyday activities. The WHO reports that 34 million children currently have some form of hearing loss (2021). Furthermore, the majority of infantile hearing impairments appear and develop in the prenatal, perinatal, and early childhood by age 3 years.

Loss of hearing during the early stages of life and before language development (*prelingual deafness*) can result in impairments, the most significant of which is linguistic deficit (Jallu et al., 2017). A child's development is particularly sensitive during the first years of life (*sensitive period*) and environmental inputs are necessary to stimulate language speaking ability and to achieve typical cognitive development (Benasich et al., 2014; Friedmann & Rusou, 2015; Litovsky, 2015; Peterson et al., 2010; Sharma et al., 2007). Individuals with hearing impairment (HI)⁵ are less exposed to natural conversations and social interactions, and impairment of communicative ability may have a serious impact on the development of social skills, relationships with peers, and overall quality of life (Abrams et al., 2016; DeCasper & Spence, 1986; Jeanes et al., 2000; Moon et al., 1993).

Neonatal hearing screenings are fundamental to identify deficits in hearing ability and initiate prompt intervention (e.g., see the Universal Newborn Hearing Screening program active in several countries including Italy⁶, the Early Hearing Detection and Intervention [EHDI], and the Joint Committee of Infant Hearing recommendations; American Academy of Pediatrics & Joint Committee on Infant Hearing, 2007). These interventions usually include speech therapy and fitting of a prosthesis (hearing aid or cochlear implant). A cochlear implant is an electronic device surgically implanted near the

⁵ I will use Hearing Loss (HL) and Hearing Impairment (HI) indistinctly.

⁶ Article 38, paragraph 2, of the Prime Minister's Decree of 12 January 2017. See also the web page of the Italian Government – Healthcare section: www.salute.gov.it/portale/temi/p2_6.jsp?lingua=italiano&id=1920&area=saluteBambino&menu=nascita

cochlea that transforms external sounds into electric pulses and transmits them to the auditory nerve to produce auditory sensation (Clark, 2015; Lenarz, 2017; Wilson & Dorman, 2008). Children implanted with the device can hear external sounds and their own voice. To optimize typical development of communicative abilities and the benefits of a cochlear implant, the EHDI has suggested guidelines based on empirical evidence:

- a) screening should be performed within the first month of life to promptly detect the hearing impairment;
- b) correct diagnosis should be confirmed within the first 3 months of life to allow initiation of clinical protocols;
- c) intervention (surgery and speech therapy) should be started by age 6 months.

These new screening techniques, treatment protocols, and diagnostic guidelines have resulted in better identification of hearing impairment and early intervention.

2.1. Communicative-pragmatic ability in children with cochlear implant

Benefits derived from the use of CI are very well known in literature (Bittencourt et al., 2012; Niparko et al., 2010; Peixoto et al., 2013). In last decades, it has been noted from several empirical research that these benefits rise when the cochlear device is implanted early. Many authors have pointed out that language acquisition is conditioned by children's early age, at the time of implantation, within a sensitive period for auditory development, as indicated by better performance in several linguistic domains – e.g., receptive vocabulary, speech perception, recognition and, production - (Baumgartner et al., 2002; Colletti et al., 2012; Dettman et al., 2007; Duchesne & Marschark, 2019; Peterson et al., 2010). Nikolopoulos et al. (2004), for instance, compared the development of grammatical abilities

in children who received a CI before the age of four and children who had the device fitted at a later age. Children were tested before receiving their CI and at two follow-up intervals, one three years after implantation and the other after five years. Those who received the implant before the age of four obtained better results than those fitted with one later; moreover, their grammatical comprehension improved with age. The authors explained this improvement as a result of the use of the CI but did not exclude an effect of the children's maturation. Geers and colleagues (2009) also investigated expressive and receptive spoken language abilities in HI children (mean age of 5 years) who received a CI before the age of 5. They reported that children's language outcomes were predicted by several factors such as the parents' education level and children's non-verbal intelligence (evaluated through language-free tasks). This result is supported by other researches showing that early interventions, among which the CI, can contribute to improve also non-verbal intelligence (Hess et al., 2014; Schlumberger et al., 2004), expanding the range of benefits provided by this device. Also, Geers and colleagues (2009) showed that the age of implantation is a significant facilitator. Globally, numerous studies underlined the key role of young age at implantation claiming that this factor predicts children's linguistic performance at a later age (Bruijnzeel et al., 2016; Ruben, 2018). For example, Yoshinaga-Itano and colleagues (2017) observed that early intervention has important benefits in terms of the development of a child's vocabulary skills, which can be comparable to those of their peers. In addition to the early practical intervention – that usually is represented by the CI implantation within 24 months of age -, children with HI generally undergo also rehabilitative speech therapies. Wie (2010) compared the performance in receptive and expressive language of children implanted between 5 and 12 months to that of children implanted between 12 and 18 months. Children implanted earlier show a better performance than children implanted later. This finding is consistent with results previously obtained by Colletti and colleagues (2009). The

authors conducted a longitudinal study on three groups of children implanted before 12 months, between 12 and 23 months and between 24 and 36 months. Data collected by Colletti et al. show that children implanted before 12 months have a typical phonological development and achieve significantly better results in linguistic tasks than children belonging to the other two groups. Overall, extensive research show that children who receive an implant before the age of 12 months have typical phonological development and achieve significantly better results in linguistic tasks than children who get the implant later.

However, while considerable research has focused on the analysis of specific aspects of language, such as phonological and morpho-syntactic skills or word recognition and spelling (Benassi et al., 2021; Caselli et al., 2012; Colin et al., 2017; Lund, 2016; Moeller et al., 2007; Pierotti et al., 2021), few studies have specifically investigated the development of the communicative-pragmatic ability in children with CIs. The development of pragmatic ability is crucial and may have several implications starting from an augmented participation and inclusion in different social contexts to increased academic success and professional attainments.

Conversational ability is the aspect that has been mainly studied (Church et al., 2017; Crowe & Dammeyer, 2021). Children fitted with CIs have been described as capable of having a conversation with peers, albeit with some difficulties. For example, Tye-Murray (2003) compared the oral conversational fluency of children with CIs to those with typical hearing, as they engaged in a conversation with a clinician, and reported that children with CIs spent more time engaging in communication breakdowns (e.g., the speaker says something but the listener does not understand) and silence than their typically hearing peers. Such dysfunctional behaviours may influence their social interactions, making them difficult and unnatural, and thus inducing other children to engage less in conversations with them. Similarly, Paatsch & Toe (2014) reported that children with HI initiate a higher percentage

of conversation topics and take longer conversation turns (i.e., words per turn) than their hearing partners. In particular, children with HI tend to ask more questions and make more personal comments (even if irrelevant). In contrast, their typically hearing cohorts use more conversational devices (e.g., “ooh”, “cool”) and are more likely to respond with short answers. This evidence suggests that children with HI might have developed these strategies in order to control the topic of the conversation. An analysis of parts of informal conversations between HI children fitted with CIs and typically hearing peers led to the conclusion that children with HI (compared to typically hearing) tend not to ask the speaker to repeat a prior utterance that they have not understood (Church et al., 2017) so that, as a result, the conversation does not flow smoothly. However, these results could also be due to the characteristics of the sample, as this included more children with a unilateral CI than with bilateral CI, or children who had not received speech therapy. Another study, conducted by Most et al. (2010), investigated the profile of pragmatic abilities among children aged between 6 and 9 years with severe hearing impairment (with CIs or hearing aids) during spontaneous conversations with an adult, and compared their performance to that of typically hearing children. The main differences characterizing the performance of children with HI are related to their inability to use verbal turn-taking consistently and appropriately, i.e., the ability to respond to the interlocutor in a timely manner and add relevant information to the conversational topic. Rinaldi et al. (2013) examined linguistic ability - production of words and sentences - and pragmatic skills - assertiveness and responsiveness in everyday dyadic interactions - in children with CIs aged between 1 and a half and 3 years. The authors used checklists and report questionnaires completed by parents to assess pragmatic abilities in children who had received the CI before the age of 12 months and those who got the implant in their second year of life and compared the performance of the two groups with normative data. Overall, the authors found a delay in the development of linguistic and pragmatic

abilities in both groups of children with a CI compared to normative data. The authors explained these differences as a result of early social experiences during child-parent interactions. Parents of children with a CI tend to be more present in the interaction by eliciting explicit requests and not giving the child enough space to propose a topic of conversation and this may lead to atypical development of pragmatic ability (DesJardin & Eisenberg, 2007). In addition, Rinaldi and colleagues did not find any differences between the group of children who received the implant within the first year and those who had it fitted between the first and the second year of life.

Another aspect that has been explored in literature is the comprehension of metaphors (Bahrami et al., 2018). Nicastrì et al. (2014) examined inferences and metaphors comprehension skills (verbal, namely the child has to explain the meaning of a metaphor that is only read by the examiner, and figurative, i.e., the child has to identify the correct metaphor among some distracting images) in children with unilateral CI aged from 6 to 15 compared to an age-matched typically hearing group. Children with hearing impairment showed substantial differences only concerning the verbal metaphors comprehension indicating that they have difficulties when reference points are absent. However, they also found that performances were correlated with age at implantation shedding light again on the importance of this factor.

As previously stated, communicative-pragmatic ability is a multimodal ability, therefore individuals can use different expressive means to communicate their intentions, and not only the linguistic one. While there are no significant differences between children with CI and with typical development in the extralinguistic ability (Ambrose, 2016), children with CI have shown difficulties in the paralinguistic ability, namely the capacity to accompany the linguistic and extralinguistic abilities with additional cues that can completely change the conveyed meaning. The paralinguistic ability comprises the capacity

to comprehend and produce emotional speech, modify the prosody, and the ability to understand and produce facial expressions. Children with CI show difficulties in the processing and recognition of prosody that can derive from their issues related to the access to the speech structure (Chin et al., 2012; Wiefferink et al., 2012). Le Maner-Idrissi and colleagues (2020) investigated the ability to process emotional prosody during speech in children with CI (age 5 to 13) considering two situations: prosody alone and prosody embedded in a situation. Results showed that, even if children with CI presented the same pattern to process emotional speech than children with typical hearing ability, their performance was poorer than the control group. Moreover, children with CI showed greater difficulty in processing prosodic cues taking simultaneously into consideration also the context. This increased complexity is explained by authors in terms of level of inferential processing. Differently than typically hearing children who based their responses principally on prosodic cues, children with CI provided answers that were more inspired by the context. Similar conclusions have been obtained by another recent study investigating emotional speech recognition (Panzeri et al., 2021).

Studies cited previously suggest that the CI is a type of intervention that can immediately ameliorate the well-being of individuals by putting them in contact with the external environment in terms of auditory perception. Improvements related to communicative abilities grow as its usage time increases providing long-term benefits. Some further studies, indeed, have shown that very early implantation of a CI, i.e., before 24 months of age, allows children with hearing impairment to develop pragmatic and social abilities comparable to those of their typically hearing peers, demonstrating that interventions based on CI can be highly promising (Guerzoni et al., 2016). Socher et al. (2019), for example, examined the occurrence of several pragmatic behaviours in everyday interactions, e.g., asking for help appropriately and responding to greetings, in school-aged

children (most of whom had received the implant before the age of 3), and compared their abilities to those of typically hearing children, finding no significant differences.

3. Study one: Pragmatic language ability in children with early cochlear implants⁷

3.1. Aims and hypothesis

The present study aims to investigate pragmatic ability in children with hearing impairment who have received an early CI intervention (i.e., before 2 years of age). More in detail, it aims to provide a comprehensive assessment of pragmatic language performance in 3 groups of school-aged children (from 6 years and 11 months old to 9 years and 11 months) with hearing impairment and a CI within the second year of life (combined with a speech therapy), compared to those of children of the same age with typical development (Control Group, CG). We hypothesized that children with CI would still exhibit difficulties in the pragmatic domain (especially the youngest group: 6;11 – 7;11). Based on findings from previous studies, these differences will be specifically observable in the ability to understand metaphors (Nicastri et al., 2014), in understanding the dialogical structure of a conversation (Paatsch & Toe, 2014; Tye-Murray, 2003) and in the ability to assume the perspective of other people in order to describe things (Peterson & Siegal, 2000; Toe & Paatsch, 2018). We also expect that age-group differences between children with CI and control group will get thinner with age. Finally, the present research will analyse the relationship between variables traditionally associated with language development in children with CIs (i.e., chronological age, gender, age at CI, and non-verbal intelligence), and the development of pragmatic skills. In detail, after controlling for the other factors (i.e., chronological age, gender and non-verbal

⁷ Portions of text and data of this chapter are published as “Hilviu, D., Parola, A., Vivaldo, S., Di Lisi, D., Consolino, P., & Bosco, F. M. (2021). Children with hearing impairment and early cochlear implant: A pragmatic assessment. *Heliyon*, 7(7), e07428.

intelligence), we expect that the age of the (the first) early CI will have a role in explaining children's pragmatic performance.

3.2. Material and methods

3.2.1. Participants

Forty-two Italian-speaking children from north-west Italy took part in the study. Eighteen children, divided into three age groups (6;11 - 7;11, 8;0 - 8;11, 9;0 - 9;11) represent the experimental group (CI, Cochlear Implant). They had severe-to-profound HI (> 70 HL dB) and had received at least one CI before the age of 24 months (see Table 1.1).

1.1 INFORMATION ABOUT PARTICIPANTS WITH HEARING LOSS AND COCHLEAR IMPLANT (CI).

| ID | Age Range (year;month) | Age in months | Gender | Age first CI in months | Age second CI in months |
|----|---------------------------|---------------|--------|---------------------------|----------------------------|
| 1 | 6;11 - 7;11 | 83 | M | 7 | 14 |
| 2 | 6;11 - 7;11 | 83 | F | 15 | 72 |
| 3 | 6;11 - 7;11 | 84 | F | 12 | 24 |
| 4 | 6;11 - 7;11 | 84 | M | 17 | 19 |
| 5 | 6;11 - 7;11 | 92 | F | 17 | 28 |
| 6 | 6;11 - 7;11 | 90 | M | 12 | 12 |
| 7 | 6;11 - 7;11 | 92 | M | 18 | 18 |
| 8 | 6;11 - 7;11 | 85 | F | 13 | 13 |
| 9 | 8;0 - 8;11 | 107 | M | 18 | 29 |
| 10 | 8;0 - 8;11 | 105 | F | 12 | 12 |
| 11 | 8;0 - 8;11 | 107 | M | 14 | 70 |
| 12 | 8;0 - 8;11 | 100 | M | 10 | 10 |
| 13 | 9;0 - 9;11 | 110 | F | 12 | 14 |
| 14 | 9;0 - 9;11 | 111 | F | 12 | 12 |
| 15 | 9;0 - 9;11 | 116 | F | 12 | 72 |
| 16 | 9;0 - 9;11 | 117 | M | 13 | 26 |
| 17 | 9;0 - 9;11 | 108 | M | 12 | 12 |
| 18 | 9;0 - 9;11 | 108 | F | 11 | 19 |

Twenty-four children with typical development, recruited from elementary schools, represent the control group (CG). The two groups of children were matched by age ($t_{(40)} = -.052$; $p = .959$), gender ($t_{(40)} = .261$; $p = .795$) and non-verbal intelligence ($t_{(40)} = -.486$; $p = .629$) (see Table 1.2 for more details on groups). Children with a CI were recruited from the ENT Department of the Martini Hospital in Turin, Italy. The presence of neuropsychiatric, neurological diseases and visual impairments – assessed by an interview of the treatment physician to children’s parents, were considered as exclusion criteria. An additional exclusion criteria was the presence of a linguistic deficit, evaluated with the Language Evaluation Battery (BVL 4-12; Marini et al., 2015), with a cut-off score of -2 SD. All children used oral language and were receiving standard Auditory-Verbal Therapy (AVT; Dornan et al., 2010). Participants and families received detailed information about the aims of the research, in accordance with the principles of the Helsinki Declaration. Both children and families were informed that participation was voluntary and that they were authorized to withdraw their participation to the research at any time. The research was approved by the Committee of Bioethics of the Azienda Ospedaliera Universitaria Città della Salute e della Scienza, Turin, Italy (Protocol number 131.410) and the children’s parents gave their informed consent.

1.2 DEMOGRAPHIC DETAILS OF THE TWO GROUPS IN THE TOTAL SAMPLE.

| | | CI | CG | <i>t</i> | <i>p-value</i> ^a |
|--|--------------|---------------|---------------|-----------------------------------|-----------------------------|
| N | 6;11 - 7;11 | 8 | 10 | | |
| | 8;0 - 8;11 | 4 | 7 | | |
| | 9;0 - 9;11 | 6 | 7 | | |
| | <i>Total</i> | 18 | 24 | | |
| Gender (F;M) | 6;11 - 7;11 | 4 F; 4 M | 6 F; 5 M | | |
| | 8;0 - 8;11 | 1 F; 3 M | 3 F; 3 M | | |
| | 9;0 - 9;11 | 4 F; 2 M | 4 F; 3 M | | |
| | <i>Total</i> | 9 F; 9 M | 13 F; 11 M | | |
| Age M(SD) | 6;11 - 7;11 | 86.75 (3.73) | 87.60 (4.95) | <i>t</i> ₍₁₆₎ = -.402 | .694 |
| | 8;0 - 8;11 | 104.75 (3.30) | 101.29 (4.46) | <i>t</i> ₍₉₎ = 1.344 | .212 |
| | 9;0 - 9;11 | 111.67 (3.83) | 113.86 (2.54) | <i>t</i> ₍₁₁₎ = -1.233 | .243 |
| | <i>Total</i> | 99.06 (12.12) | 99.25 (11.90) | <i>t</i> ₍₄₀₎ = -.052 | .959 |
| Non-verbal intelligence CPM | 6;11 - 7;11 | 23.62 (2.56) | 23.70 (5.29) | <i>t</i> ₍₁₆₎ = -.037 | .971 |
| | 8;0 - 8;11 | 29.50 (5.92) | 28.00 (3.60) | <i>t</i> ₍₉₎ = .531 | .608 |
| | 9;0 - 9;11 | 29.16 (3.12) | 32.57 (1.62) | <i>t</i> ₍₁₁₎ = -2.526 | .028 |
| | <i>Total</i> | 26.78 (4.49) | 27.54 (5.40) | <i>t</i> ₍₄₀₎ = -.486 | .629 |

^a Statistically significant p-values (< .05) are shown in bold.

Abbreviations: CI = Cochlear Implant group; CG = Control group; M = mean; SD = standard deviation; F = female; M = male.

3.2.2. Material

Pragmatic ability assessment

We administered the *Language Pragmatic Abilities - APL Medea* (Lorusso, 2009), a validated battery developed to evaluate the comprehension and use of verbal language in children ranging in age from 5 to 14 years (normative data for the Italian population are also available for different age ranges). The battery has already been used to assess pragmatic skills in children and young adolescents with CI (Nicastri et al., 2014) and in other clinical populations (Cardillo et al., 2018, 2021). All tasks in the APL Medea battery are presented to children in the form of game.

The battery consists of five tasks:

1. *Metaphors* (M): this task (preceded by a practice trial) evaluates the ability to understand the implicit meaning of sentences going beyond the literal meaning. Eight familiar metaphors are orally presented to the child (i.e., read by the examiner), who has to understand the meaning of each. For example: the examiner reads “Today Mark feels like a lion” (in Italian “Oggi Marco si sente un leone”) and the child has to explain that this is a way to say that Marco feels very strong (the maximum score for this task is 16)⁸;
2. *Implicit meaning comprehension* (IMC): evaluates the ability to make inferences on implicit contents about situations. It comprises three short stories that are read to the participant, followed by a set of questions designed to investigate the correct understanding of the situation described in the story. For instance: “My arm hurts a lot” says Michael while lying on the examination table. “Do not worry, now I am going to give you a drug that will make disappear the pain” answers Robert. Subjects are asked then questions about the context where the scene is happening, who are the characters, and which is Robert’s profession (the maximum score for this task is 14);
3. *Comics* (C): evaluates the ability to understand conversations in terms of their dialogical structure. The examiner reads four pieces of comics that are incomplete and the child is asked to complete them. For example, in one comic there is a chick asking to his mother “Mom, can I go outside?” and his mother answers “Yes, but do not be late”. After that, the chick meets a kitten and says “Finally we can see each other!” and the kitten answers “Yes, I wanted to see you!”. Then, in the last scene, the chick says something (his speech bubble is empty) and the kitten answers “We can play hide and seek!” (the maximum score for this task is 12);

⁸ It should be noted that the task *Metaphors* of the APL Medea comprises some items that are idioms.

4. *Situations (S)*: evaluates the understanding of sentences considering the relative context. In this task, the examiner reads some stories and then asks the child a simple question to test his or her understanding, or presents some options and the child is invited to choose the most appropriate in relation to a specific context. For instance: the teacher says to John “How is that possible that you are always late?”. The child is then questioned “What do you think John will answer?” (the maximum score for this task is 11);
5. *Colours Game (CsG)*: evaluates the ability to understand other people’s mental representations. The examiner presents the child with a game, explains the rules, and describes the material needed. The game is very simple: there is a six-sided dice and in each side of the dice there is a coloured dot (2 sides have a yellow dot, one side a green dot, one side a red dot and one side a black dot) or a symbol (a smiley face), two pawns and a cardboard divided into three coloured horizontal-sections (starting from below: yellow, green and red). One of the players rolls the dice and based on the result (a colour or the symbol) the player can put the pawn in the respective coloured section of the cardboard (the black dot indicates that the player must stop for a turn, while the smiley face means that the player can roll the dice twice). The first who reaches the red section of the cardboard wins the game. The examiner and the child play a few matches to make sure the child has understood correctly. The participant is then asked to describe the game to another child who was not present before and who will listen to the recording afterwards without knowing the rules or the material that is needed. The description is then transcribed and analysed, i.e., the child has to report a series of elements such as needed material, game rules, meaning of the smiley face on the dice, etc. (the maximum score for this task is 15).

The final maximum score of the APL Medea is 68.

Non-verbal intelligence assessment

To evaluate the role of non-verbal intelligence in pragmatic performance, we administered the Coloured Progressive Matrices (CPM; Raven, 1947). Children were asked to solve 36 coloured puzzles (equally divided into three sets: A, AB, B), by choosing the compatible missing part from six items (five of which are distractors). Each correct answer is given one point. The maximum score for each set is 12 and the total maximum score is 36. No difference was found between the two groups.

3.2.3. Procedure

Research assistants with a degree in Psychology administered the two tests (APL Medea and CPM) in one single session, lasting approximately 40 minutes. All children of both groups (CI and CG) were tested individually (only the examiner was in the same room with the participant). Children with CIs were tested at the hospital in a room already familiar to them while children with typical hearing abilities were evaluated at school, in an empty classroom. The APL Medea was presented to both groups in an oral form (the examiner read all items) with the support of material presented in paper format, i.e., drawings. In case children did not understand or hear an instruction, the examiner repeated it. If children appeared tired or distracted, the examiner proposed them a short pause (e.g., asking them if they wanted a glass of water).

3.2.4. Data analyses

Analyses were performed with IBM SPSS Statistics 26 software.

Pragmatic ability assessment

To investigate the differences in pragmatic performance between the two groups of children (CI vs CG), and how these differences are associated with the children's age group and the type of pragmatic task, we submitted their APL Medea scores to a $2 \times 3 \times 5$ repeated measures ANCOVA, with Group (CI vs CG) and Age (6;11 – 7;11 vs 8;0 – 8;11 vs 9;0 – 9;11) as *between-subjects factors*, and Tasks (Metaphors, Implicit Meaning Comprehension, Comics, Situations and Colours Game) as the *within-subjects factor*. Since we were interested in pragmatic performance, we inserted the CPM raw score as a covariate in order to control for the role of non-verbal intelligence, which is generally a predictor of language ability (Geers et al., 2009; Marini et al., 2008).

Role of age, gender, non-verbal intelligence, and age at implantation

To investigate if the age at implantation is a significant predictor for children's general pragmatic ability (APL Medea total score), we ran a hierarchical linear regression analysis on the children in the CI group. Specifically, we controlled for the role of age, gender and Raven's CPM score (Raven, 1947), and entered these in the first step of the model. Therefore, children's age at implantation was entered as independent variable in the second step in order to verify whether, keeping under control the previous variables, it represents a significant predictor of pragmatic ability in children with a CI.

3.3. Results

3.3.1. Pragmatic ability assessment

Individual performance at the APL Medea tasks of children with CI are presented in Table 1.3.

1.3 INDIVIDUAL PERFORMANCE OF CHILDREN WITH CI AT EACH TASK OF APL MEDEA BATTERY.

| ID | Metaphors | IMC | Comics | Situations | Colors Game | Total APL Medea |
|----|-----------|-------|--------|------------|-------------|-----------------|
| 1 | 2.00 | 6.50 | 1.00 | 5.00 | 4.00 | 18.50 |
| 2 | 4.00 | 1.00 | 2.00 | 6.00 | 4.00 | 17.00 |
| 3 | 6.00 | 3.00 | 6.00 | 4.00 | 7.00 | 26.00 |
| 4 | 3.00 | 4.00 | 6.00 | 1.00 | 8.00 | 22.00 |
| 5 | 2.00 | 4.50 | .00 | 2.00 | 2.00 | 10.50 |
| 6 | .00 | 8.00 | 1.00 | 1.00 | 8.00 | 18.00 |
| 7 | 3.00 | 4.50 | .00 | 3.00 | 8.00 | 18.50 |
| 8 | 6.00 | 6.00 | 9.00 | 4.00 | 6.00 | 31.00 |
| 9 | 7.00 | 6.00 | 2.00 | 3.00 | 10.00 | 28.00 |
| 10 | 4.00 | 5.50 | 4.00 | 2.00 | 4.00 | 19.50 |
| 11 | 9.00 | 9.00 | 5.00 | 4.00 | 10.00 | 37.00 |
| 12 | 8.00 | 9.50 | 5.00 | 6.00 | 10.00 | 38.50 |
| 13 | 10.00 | 12.50 | 9.00 | 8.00 | 10.00 | 49.50 |
| 14 | .00 | 5.00 | 6.00 | 7.00 | 8.00 | 26.00 |
| 15 | 4.00 | 10.50 | 10.00 | 8.00 | 7.00 | 39.50 |
| 16 | 8.00 | 11.00 | 10.00 | 9.00 | 11.00 | 49.00 |
| 17 | 8.00 | 6.50 | 8.00 | 4.00 | 9.00 | 35.50 |
| 18 | 2.00 | 9.00 | 10.00 | 2.00 | 9.00 | 32.00 |

The repeated measures ANCOVA showed a main effect of Group ($F_{(1,35)} = 8.723$; $p = .006$; $\eta^2_p = .200$), indicating that participants in the CI group generally performed significantly worse than those in the CG, and a main effect of Age ($F_{(2,35)} = 5.1566$; $p = .008$; $\eta^2_p = .241$), indicating that performance improves with age. The effect of APL Medea tasks was not significant ($F_{(4,140)} = .288$; $p = .885$; $\eta^2_p = .008$). The interaction Group*Age*Tasks was significant ($F_{(8,140)} = 2.922$; $p = .005$; $\eta^2_p = .143$) indicating that children's performance on the Medea tasks depends on both the Group and the Age of participants. These effects were significant after controlling for the role of the covariate non-verbal intelligence, i.e., Raven's CPM.

In order to explore the significant interaction Group*Age*Tasks, we performed pairwise comparisons with Bonferroni correction. Considering the different Age groups (three levels: 6;11 – 7;11 vs 8;0 – 8;11 vs 9;0 – 9; 11), pairwise comparisons revealed that

only children with a CI in the youngest age group (6;11 – 7;11) significantly differed from their hearing peers in terms of overall performance in overall APL Medea tasks ($p = .012$) (see Figure 1.1).

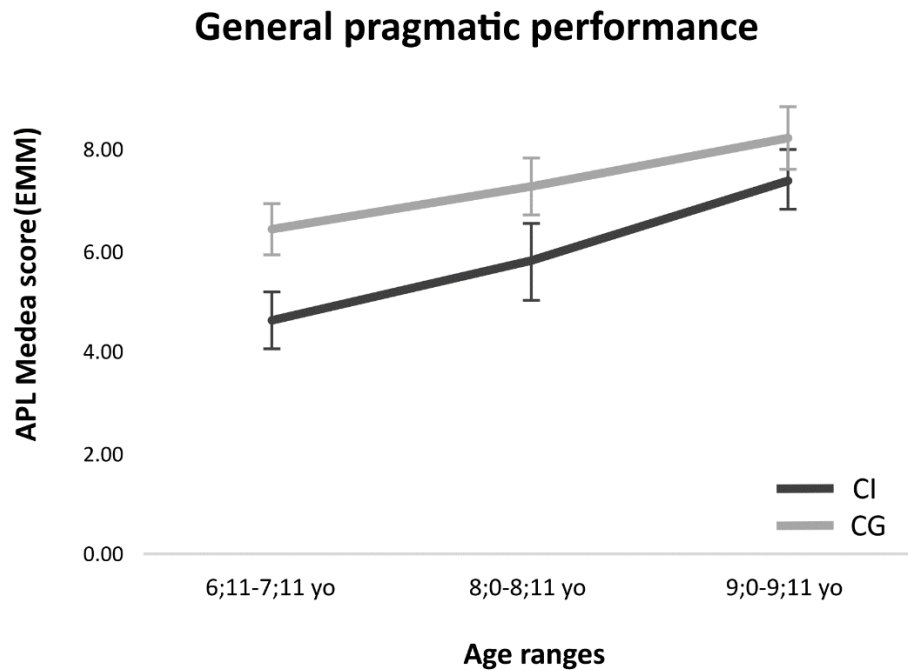


FIGURE 1.1 PRAGMATIC PERFORMANCE. GENERAL PRAGMATIC PERFORMANCE (APL MEDEA SCORE) ACROSS THE DIFFERENT AGE RANGES IN THE GROUP OF CHILDREN WITH A COCHLEAR IMPLANT (DARK GREY) AND THE CONTROL GROUP (LIGHT GREY). PAIRWISE COMPARISONS REVEALED THAT THE ONLY SIGNIFICANT DIFFERENCE WAS FOUND IN THE 6;11 – 7;11 AGE RANGE BETWEEN THE COCHLEAR IMPLANT GROUP AND TYPICALLY HEARING CHILDREN ($p = .012$). N.B. COVARIATES INCLUDED IN THE MODEL WERE EVALUATED ON THE FOLLOWING VALUE: RAVEN’S CPM = 27.214. ERROR BARS INDICATE STANDARD ERRORS.

Considering separately the different Medea tasks (Metaphors, Implicit Meaning Comprehension, Comics, Situations, and Colours Game), children with CIs only differed from their hearing peers (all age groups pooled together) in the Comics task ($p = .008$) and Colours Game task ($p = .018$) (see Table 1.4).

1.4 MEANS (STANDARD DEVIATIONS) OF THE TWO GROUPS ON EACH APL MEDEA TASK AND TOTAL SCORE.

| | CI, N = 18 | | CG, N = 24 | | p-value ^a |
|--------------|---------------|---------------|------------|---------------|-------------------------|
| | Min - Max | M (SD) | Min - Max | M (SD) | |
| Metaphors | 0 - 10 | 4.78 (3.08) | 2 - 13 | 6.42 (2.70) | .080 |
| IMC | 1 - 12.50 | 6.78 (3.02) | 2 - 12.50 | 8.25 (2.95) | .124 |
| Comics | 0 - 10 | 5.22 (3.59) | 1 - 12 | 7.00 (2.72) | .008 |
| Situations | 1 - 9 | 4.39 (2.48) | 1 - 8 | 4.96 (1.68) | .431 |
| Colors Game | 2 - 11 | 7.50 (2.57) | 2 - 15 | 9.67 (3.06) | .018 |
| <i>Total</i> | 10.50 - 49.50 | 28.67 (11.18) | 14 - 54 | 36.29 (10.05) | .006^b |

^a Statistically significant p-values (< .05) adapted for multiple comparisons with Bonferroni are shown in bold.

^b Significance level resulting from the ANCOVA analysis.

P-values indicate the level of significance in pairwise comparisons between the two groups on APL Medea tasks. Abbreviations: CI = Cochlear Implant group; CG = Control group; M = mean; SD = standard deviation; IMC = Implicit Meaning Comprehension.

Finally, considering the different Tasks in the different Age groups, we found that children with a CI differed from their hearing peers in the Age Group 6;11 – 7;11 years in the Colours Game task ($p = .046$), in the Age Group 8;0 - 8;11 in the Comics task ($p = .000$), and in the Age Group 9;0 – 9;11 in the Metaphors task ($p = .014$) (see Figure 1.2). Overall, pairwise comparisons indicated that the difference between children with a CI and their hearing peers was greater in the Age group 6;11 – 7;11, but tended to be smaller in the other two groups (8;0 – 8;11 and 9;0 – 9;11). The Medea tasks in which children in the CI group were most impaired compared to the hearing group were Comics and the Colours Game (see Figure 1.2).

Finally, the effect of CPM was also significant ($F_{(1,35)} = 6.513$; $p = .015$; $\eta^2_p = .157$) indicating an association between non-verbal intelligence and children's general pragmatic performance (both groups, i.e., CI and CG, pooled together).

Participants performance on APL Medea's tasks

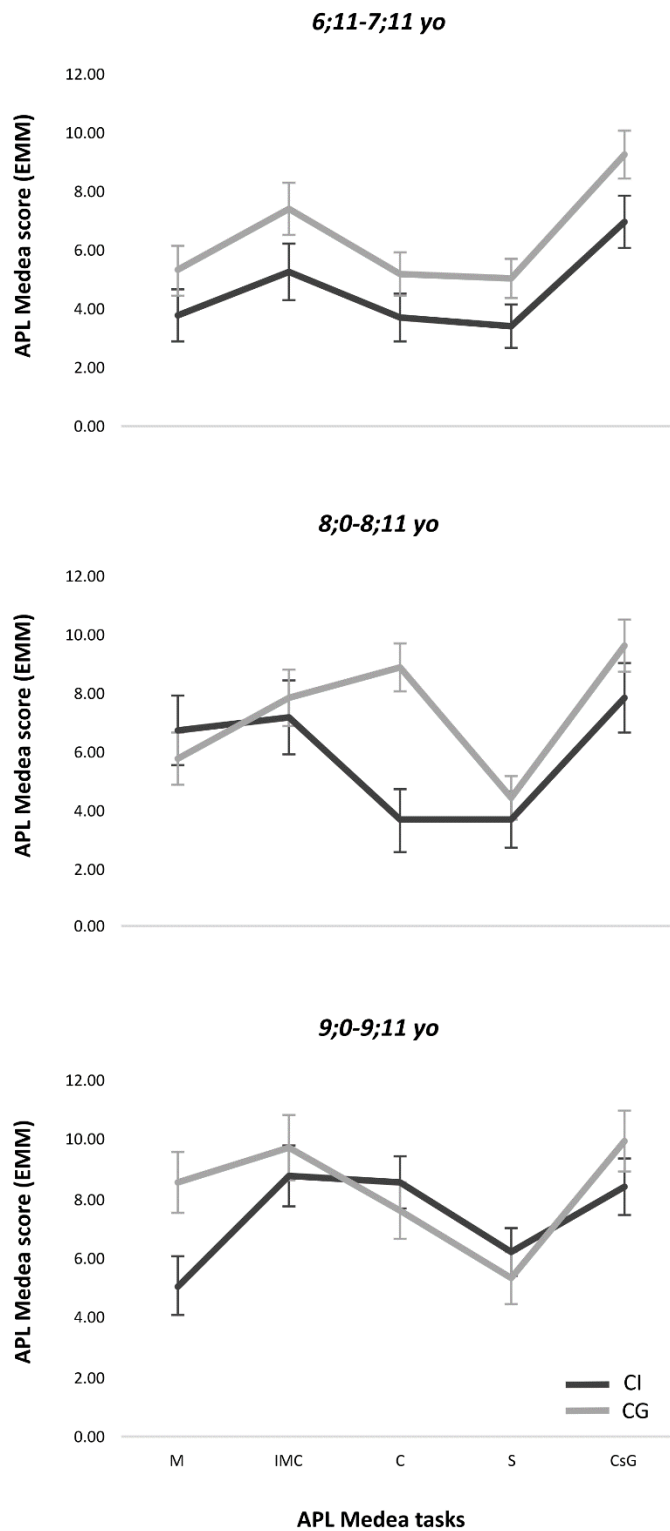


FIGURE 1.2 PRAGMATIC PERFORMANCE IN DIFFERENT AGE-RANGES. SCORES ON APL MEDEA TASKS ACROSS THE DIFFERENT AGE RANGES IN THE GROUP OF CHILDREN WITH A COCHLEAR IMPLANT (DARK GREY) AND THE CONTROL GROUP (LIGHT GREY). ABBREVIATIONS: M = METAPHORS; IMC = IMPLICIT MEANING COMPREHENSION; C = COMICS; S =SITUATIONS; CsG = COLORS GAME. ERROR BARS INDICATE STANDARD ERRORS.

3.3.2. Role of age, gender, non-verbal intelligence, and age at implantation at implantation

The hierarchical linear regression analysis showed a significant impact of age and CPM (see Table 1.5). More in detail, the R^2 value indicates that age, gender and CPM account for 60% of the variation in pragmatic performance. *Age at implantation* was found to be a significant predictor too: the R^2 indicates that it accounts for an additional 12% of the variation in pragmatic performance ($\beta = -.370$; $t = - 2.363$; $p = .034$).

1.5 HIERARCHICAL LINEAR REGRESSION ANALYSIS OF CHILDREN FITTED WITH A COCHLEAR IMPLANT WITHIN 24 MONTHS OF AGE (N = 18).

| Dependent Variable | Predictors | R^2_{Adj} | R^2_{Change} | F_{Change} | Sig. F_{Change} |
|--|----------------|-------------|----------------|--------------|-------------------|
| Pragmatic ability (Tot. score APL Medea) | Model 1 | .514 | .600 | 6.990 | .004 |
| | Model 2 | .634 | .120 | 5.582 | .034 |

Model 1 (age, gender, and non-verbal intelligence measured by CPM); Model 2 (age at implantation).

3.4. Discussion

The present research aimed to study pragmatic ability in school-aged children with a cochlear implant fitted within the first 24 months of life. Firstly, we analysed their performance on different pragmatic tasks and compared them to a group of typically hearing peers. Overall, children with a CI obtained lower scores in the total of the APL Medea battery compared to typically hearing children. Furthermore, once controlled the role of non-verbal intelligence, the performance of children with CI increased with age.

In order to deeper investigate the role of chronological age and the differences in pragmatic performance between children with a CI and typically hearing peers, we performed a fine-grained analysis across the different age groups (7-, 8- and 9-year-olds).

Results indicated that the differences between children with a CI and typically hearing peers in overall pragmatic performance varied across age groups. Indeed, only the younger age group (i.e., 6;11–7;11 age range) showed a significant difference while the two older groups (8;0–8;11 and 9;0–9;11) performed equally to controls. The 8- and 9-year-old children showed substantial differences only in specific tasks, i.e., in the Comics task in the group of 8-year-olds and in the Metaphors task in the 9-year-old age group. This result suggests that since the difference in performance is greater in younger children (7-year-olds), it seems to decrease as age increases. A possible explanation for this result is the major exposure, in terms of time, to the auditory stimuli experienced by older children (see Duchesne & Marschark, 2019; Marschark et al., 2019).

By focusing on the single tasks investigated by the APL Medea - Metaphors, Implicit meaning comprehension, Comics, Situations and Colours Game - the overall performance of children with a CI, was significantly poorer, with respect to the CG, in two tasks only: Comics and Colours Game.

The Comics task evaluates the ability to use pragmatics appropriately in a context, to understand a conversation and to respect its dialogical structure (Lorusso, 2009). Our finding is consistent with previous studies showing that children with a CI have difficulties in mastering a conversation, turn-taking and in recovering from communicative breakdowns (Paatsch & Toe, 2014; Tye-Murray, 2003). We believe this lower level of ability may be the consequence of reduced exposure of children with HI to different communicative contexts and partners due to their condition. This may result in difficulty in adopting the appropriate strategy to manage a conversation correctly. Moreover, as other authors have pointed out, this difficulty may also be the result of an inappropriate strategy adopted by this population aimed at controlling conversational turns when they are engaged in conversations (Church et al., 2017; Toe & Paatsch, 2013). This control, which consists, for instance, in choosing

the discourse topic or frequently not leaving space for the interlocutor to intervene, helps this population to reduce communicative breakdowns and limit the overuse of communication failure recovery strategies, that could make the linguistic interaction unnatural. An alternative explanation to the delay in mastering conversations could be ascribed to the delay in language development, e.g., vocabulary, as already highlighted by previous research (Rinaldi et al., 2013). The delayed exposure to auditory stimuli can cause a delay in the linguistic acquisition, which in turn supports the pragmatic ability.

The second significant difference we found concerns the Colours Game task, that evaluates children's ability to assume another person's perspective. In literature, this ability is usually connected with the concept of Theory of Mind (ToM; Premack & Woodruff, 1978), i.e., the ability to attribute mental states to the self and to others (Harwood & Farrar, 2006; Taylor et al., 1991). Studies in the literature showed that ToM develops beyond childhood, through adolescence and up to the adulthood (Blakemore, 2008; Brizio et al., 2015). The literature also pointed out a delay in the development of ToM in children with HI (Peterson & Siegal, 2000; Russell et al., 1998). A correlation between pragmatics and ToM is also well documented in the literature (Sperber & Wilson 2002; Nilsen et al., 2011; Tirassa & Bosco, 2008), even if these two cognitive abilities do not completely overlap (Arcara & Bambini, 2016; Bosco & Gabbatore, 2017a, 2017b; Bosco, Tirassa, et al., 2018). Our findings are thus in line with previous studies showing a delay in the developmental trend of ToM in children with HI (Matthews et al., 2018; Meristo & Hjelmquist, 2009; Most et al., 2010). This finding is also in line with previous studies investigating children's expository discourse, i.e., the use of language to describe something or an event (Toe & Paatsch, 2018). The Colours Game task, in this sense, can be considered an example of expository discourse since it requires the child to describe the rules of a game.

A detailed analysis also revealed that the Metaphors task differed significantly only in the 9-year-old children. This task evaluates the ability to go beyond the literal meaning and understand what the partner is really communicating. Despite being familiar metaphors⁹, our findings are in line with the results of a previous study by Nicastrì et al. (2014) showing a difference between children with a CI and hearing peers in the same task. Furthermore, the literature suggests that the ability to understand metaphors develops fully in late childhood (Billow, 1975; Winner et al., 1976). Therefore, it is plausible to assume that the difference we detected in our results is due to a delay in the development of this ability compared to typically hearing peers, rather than a deficit. Additionally, understanding metaphors seems to be linked to other abilities, such as ToM (Lecce et al., 2019), that we also found to be delayed, in a certain way, in our sample of children with a CI.

Finally, we also found a role for the covariate non-verbal intelligence in overall pragmatic performance, in line with previous literature showing that non-verbal intelligence has a significant role in language acquisition (Geers et al., 2009). Despite the important role of IQ, the difference in pragmatic performance between children in the CI group and normal hearing peers is still significant, thus indicating that the pragmatic impairment is specific and not only attributable to the role of non-verbal intelligence.

A further aim of the present study was to investigate the role of age at implantation in pragmatic performance. In our model we first assessed the role of non-verbal intelligence and age as predictors of pragmatic performance, and then added age of implantation as predictor in the second model, to test whether this provides an independent contribution to explain pragmatic performance of children with a CI. Age at implantation was found to be a moderate but significant predictor, confirming the hypothesis that an early CI, as a result of

⁹ As previously noted, the *Metaphors* task also included idioms that could have somehow influenced our result. Past studies have shown that young children, i.e., 7-8 years old, are more likely to interpret idioms literally while older children, i.e., 9-10 years old, are more able to grasp the figurative meaning (Ackerman, 1982; Levorato & Cacciari, 1995).

an early intervention, contributes to an improvement in pragmatic ability. Although the time window for CI implantation used in our study was quite short (24 months), it was still possible to observe differences in pragmatic performance depending also on the age of implantation. This result is in line with other findings that underlined the association between age at implantation and pragmatic performance (Guerzoni et al., 2016; Nicastrì et al., 2014), but is in contrast with the study by Socher et al. (2019). However, this difference may be due to various variables, such as the data collection procedure. In the study by Socher et al. (2019), in fact, pragmatic measures were provided by parents using rating scales for each pragmatic behaviour occurrence and this could have somehow influenced their evaluations.

Our result is encouraging and support previous findings available in the current literature (Guerzoni et al., 2016; Nicastrì et al., 2014) that early diagnosis together with concrete support in the form of a hearing device in children who have subsequently also undergone a speech treatment, will help them to limit their pragmatic difficulties in a later age of development.

There are several limitations to the present study. Firstly, the research included a small sample of children with a CI and moreover, the children were not equally divided across age ranges. This might have affected our results, since almost half of the sample were less than 8 years old. A larger and more balanced sample might have helped us to underline differences or similarities between the two groups. Secondly, since this was not a longitudinal study, we were only able to make assumptions about the developmental trend in the pragmatic ability of children with a CI. Future research should investigate pragmatic performance in the same group of children at different time intervals. Thirdly, sociodemographic information was not collected and from previous literature it is well known that several elements intervene during a child's development, such as socioeconomic status or the parents' educational level (Geers et al., 2009) and the presence of siblings

(McAlister & Peterson, 2013; Woolfe, 2003). Finally, we did not collect measures on vocabulary, verbal fluency or verbal working-memory, skills that previous literature has shown to be correlated to the pragmatic one (Kronenberger et al., 2018; Marshall et al., 2015; Matthews et al., 2018).

Our findings provide evidence in favour of early CI implantation in children with HI. However, early implantation alone is not sufficient to assure typical pragmatic ability development. Our results support findings from previous studies (Guerzoni et al., 2016; Nicastrì et al., 2014) providing a more comprehensive evaluation of pragmatics. Globally considered, the existing research suggest that along with early prosthesis, and early verbal and speech treatment (Binos et al., 2021), specialists could also enrich interventions to include programs specifically focused on pragmatic abilities. Indeed, a previous research has shown that the Auditory-Verbal Therapy does not provide a direct effect in improving pragmatic development (Toe et al., 2016). Therefore, programs should be expanded to cover other aspects in which children with a CI appear to have difficulties, such as conversation, perspective-taking, figurative language, and its affective aspects (for instance, in the case of irony and sarcasm), and also to the argumentative abilities needed for both comprehension and production of figurative language in the developmental age (see Cocco & Ervas, 2021 for a review). Overall, these difficulties have several implications for children with a CI, such as influencing their social interactions by inducing other individuals to interact less with them (Cawthon et al., 2015). Therefore, early conversations with adults (Meristo et al., 2016) and peers are vital for children because in this way they experience natural and diversified forms of interactions and are able to practice pragmatic skills in different contexts. Finally, future research should also include the investigation of other cognitive aspects, as for example ToM, working memory and other expressive means, as non-verbal

and paralinguistic ones, in order to understand deeper communicative difficulties in children with CI.

4. Study two: Multimodal pragmatic abilities in children with early cochlear implants¹⁰

4.1. Aims and hypothesis

In the present study we aimed to provide a comprehensive assessment of communicative-pragmatic ability in children with CIs and typically hearing peers from a multimodal point of view. We used the Assessment Battery for Communication (ABaCo), a tool for investigating a wide range of communicative aspects expressed through different expressive modalities, validated on the Italian adult population (Angeleri et al., 2012) and adapted for children (Bosco et al., 2012; Sacco et al., 2008). ABaCo provides a complete evaluation of communicative-pragmatic abilities in children with typical (Bosco et al., 2013; Bosco & Gabbatore, 2017a) and atypical development (Angeleri et al., 2016), and in adult clinical populations (Bosco, Parola, Sacco, et al., 2017; Parola et al., 2016). Specifically, we investigated different expressive means, i.e., linguistic, non-verbal/extralinguistic (e.g., gestural), paralinguistic (e.g., prosodic), in addition to sensitivity to the social context and conversational skills. Further, we wanted to investigate the development of communicative-pragmatic ability across different age groups, i.e., 6;11 – 7;11; 8;0 – 8;11; 9;0 – 9;11, and assess whether the differences between children with CI and typically hearing (TH) peers vary as a function of children's age. Moreover, since the previous literature has shown that non-verbal intelligence has a significant role in language acquisition (Watson et al., 1982), we wanted to control for the effect of non-verbal intelligence in pragmatic performance, in order to rule out its role in explaining our results.

¹⁰ Portions of text and data of this chapter are currently submitted and under review as “Parola, A., Hilviu, D., Vivaldo, S., Di Lisi, D., Consolino, P., & Bosco, F. M. “Development of communicative-pragmatic abilities in children with early cochlear implant” to the *Journal of Child Language*.

Finally, we aimed to investigate whether several factors (i.e., age of first CI, age of second CI and non-verbal intelligence) influence the development of communicative-pragmatic abilities. Our sample thus included children who had undergone bilateral cochlear implantation at different ages -, and we examined whether the age of first and second implantation could predict the development of pragmatic abilities during the early school years. We made the following hypotheses:

- I. We expect to observe significant differences in overall communicative-pragmatic performance between children with CI and TH peers on the ABaCo. Specifically, we expect these differences to be greater between children with CI and TH peers in the youngest age groups, i.e., we hypothesize a greater improvement in pragmatic ability in children with CI with their increase in age, compared to the TH peers, given a longer exposure to acoustic and social communicative stimuli.
- II. After controlling for the other variables (e.g., age, age of second implantation, non-verbal intelligence), we predict an effect of age of first implantation on pragmatic development.

4.2. Methods

4.2.1 Participants

Forty-four children were included in the present study. Twenty-two children (aged between 6;11 years and 9;11 years; 12 females and 10 males; mean age 98.64 ± 11.19 months) were diagnosed with profound-to-severe hearing impairment (> 70 dB loss). All children with HI had been fitted with bilateral CIs. See Table 2.1 for more details on the experimental group, and for the age of first and second implantation for each participant. The children were recruited from the ENT Department of the Martini Hospital in Turin, Italy. Exclusion criteria for children with CIs were the presence of neurological disease or

neuropsychiatric illness, visual and language impairments. While the first three were eventually reported by the family pediatrician, the language impairments were assessed with the Language Evaluation Battery (BVL 4-12; Marini et al., 2015). The cut-off score was set at -2 SD. All children had been through a treatment program, i.e., auditory/verbal program and used oral language. No data related to the use of sign language was collected.

The control sample comprised 22 children with typical hearing development (TH) recruited from local elementary schools (aged between 6;11 years and 9;11 years; 12 females, 10 males; mean age 100.00 ± 12.25 months). All children were native Italian speakers. No significant difference was observed between the group with CIs and the group of typically hearing children in non-verbal intelligence ($t_{(42)} = -.46$ $p = .65$) measured with Raven's coloured progressive matrices (CPM; Raven, 1947).

Both families and children received detailed information about the aims of the research as suggested by the principles of the Helsinki Declaration. Parents or caregivers had to sign the informed consent form, and gave permission for the sessions to be video-recorded. The children and their parents were informed that the participation to the study was voluntary and they could interrupt the test at any time without having to give a reason. The research was approved by the Committee of Bioethics of the Azienda Ospedaliera Universitaria Città della Salute e della Scienza, Turin, Italy (Protocol number 131.410).

TABLE 2.1 INFORMATION ON THE EXPERIMENTAL SAMPLE: CHILDREN WITH HEARING IMPAIRMENT AND BILATERAL COCHLEAR IMPLANTS (CI). AGE RANGE COLUMN INDICATES AGE IN YEARS AND MONTHS.

| ID | Age Range | Age in months | Gender | Age left CI in months | Age right CI in months |
|----|-------------|---------------|--------|-----------------------|------------------------|
| 1 | 6;11 - 7;11 | 83 | M | 7 | 14 |
| 2 | 6;11 - 7;11 | 83 | F | 72 | 15 |
| 3 | 6;11 - 7;11 | 84 | F | 24 | 12 |
| 4 | 6;11 - 7;11 | 84 | M | 17 | 19 |
| 5 | 6;11 - 7;11 | 90 | F | 38 | 30 |
| 6 | 6;11 - 7;11 | 85 | F | 13 | 13 |
| 7 | 6;11 - 7;11 | 90 | M | 12 | 12 |
| 8 | 6;11 - 7;11 | 91 | F | 96 | 84 |
| 9 | 6;11 - 7;11 | 92 | F | 17 | 28 |
| 10 | 6;11 - 7;11 | 92 | M | 18 | 18 |
| 11 | 8;0 - 8;11 | 100 | M | 10 | 10 |
| 12 | 8;0 - 8;11 | 105 | M | 30 | 30 |
| 13 | 8;0 - 8;11 | 105 | F | 12 | 12 |
| 14 | 8;0 - 8;11 | 107 | M | 29 | 18 |
| 15 | 8;0 - 8;11 | 107 | F | 121 | 60 |
| 16 | 8;0 - 8;11 | 107 | M | 70 | 14 |
| 17 | 9;0 - 9;11 | 108 | M | 12 | 12 |
| 18 | 9;0 - 9;11 | 108 | F | 19 | 11 |
| 19 | 9;0 - 9;11 | 110 | F | 14 | 12 |
| 20 | 9;0 - 9;11 | 111 | F | 12 | 12 |
| 21 | 9;0 - 9;11 | 112 | M | 27 | 27 |
| 22 | 9;0 - 9;11 | 116 | F | 72 | 12 |

4.2.2. Material

Assessment of communicative-pragmatic ability

All the children were assessed using the Assessment Battery for Communication (ABaCo; Angeleri et al., 2012; Bosco et al., 2012; Sacco et al., 2008). The battery is comprised of five scales (linguistic, extralinguistic, paralinguistic, contextual and conversational), each of which is designed to evaluate a different aspect of communication with regard to comprehension and production. The protocol is made up of 172 items, which consist of videotaped scenes shown to the participants and face-to-face interactions with the

experimenter. Participants are asked to answer some questions to show their understanding of the situation, or to complete an interaction. The instrument is designed for Italian native speakers. Subjects' performance is coded off-line by two independent judges who examine the recordings and are not informed about the aim of the research or identity of the participants. All items are evaluated as correct (1 point) or incorrect (0 points). The degree of reliability between the two judges was calculated using 28.57% of the sample and the average ICC measure was .83 with a 95% confidence interval from .77 to .88 ($p < .001$).

The following sections provide a detailed description of each scale (for examples of each scale see Appendix A). It is worth noticing that the majority of the scales of ABaCo focuses mainly on the evaluation of one expressive modality, however some of them include the evaluation at the same time of the interaction of more than one expressive mean (e.g., the linguistic scale, previously called ecological scale, evaluates the use of language but includes also the use of facial expressions and changes in the tone of voice).

Linguistic scale

This scale focuses on linguistic aspects of communication and, in particular, it evaluates the comprehension and production of different communicative acts, such as basic communicative acts i.e., statements, questions, requests and commands (Kasher, 1994), standard communicative acts, irony and deceit, using language (Searle, 1975).

- **Comprehension:** in the case of basic communicative acts, the subject is asked to evaluate the truthfulness of a statement, answer a simple question, perform a requested action, or carry out an order. In the case of standard communicative acts, irony and deceit, the subject watches some short videos in which an actor asks the partner a question and the partner answers. The subject has to demonstrate an understanding of the situation and of the answer, by simply answering some questions about the scene.

- **Production:** in the case of basic communicative acts, the subject is required to produce a statement, a question, a request, or a command. In the case of standard communicative acts, irony and deceit, the subjects watch some videos in which an actor formulates a question depending on the context and the subject is asked to answer the question.

Extralinguistic scale

This scale assesses the comprehension and production of communicative acts (i.e., basic communicative acts, standard communicative acts, irony and deceit) using gestures. The tasks are similar to those of the linguistic scale, except for the use of language.

- **Comprehension:** the subject is required to show an understanding of communicative acts expressed through gestures.
- **Production:** the subject is required to produce communicative acts using gestures only.

Paralinguistic scale

The scale evaluates the comprehension and production of all non-verbal cues (e.g., prosody, proxemics) that supplement meaning expressed through linguistic and extralinguistic modalities.

- **Comprehension:** this subscale includes three types of tasks (i.e., basic emotions, basic communicative acts and paralinguistic incongruity). For basic communicative acts and communicative acts that express emotions, video clips in which an actor speaks an invented language are shown to the subject and he/she is asked to recognize the expressed emotion or to demonstrate comprehension of the basic communicative act that is produced. As far as paralinguistic incongruity is concerned, the participant watches videos in which an actor asks his interlocutor a question, and the latter answers with a

sentence that is appropriate to the specific situation, but in which the paralinguistic indicators are contradictory. The subject has to recognize the paralinguistic incongruity.

- **Production:** this comprises two tasks, i.e., basic communicative acts that express emotions, and basic communicative acts. The subject is asked to produce communicative acts using appropriate paralinguistic aspects, in order to express a basic emotion, a statement, a question, a request or a command.

Contextual scale

This scale assesses the child's ability to use appropriate communicative behavior with respect to different social contexts.

- **Comprehension:** the subject has to demonstrate his/her knowledge of the rules that guide communicative interaction in a specific social context. This ability is assessed through two tasks: violation of the norms of discourse and violation of social norms. In the first case, the subject must recognize the violation of a Grice's maxim (Grice, 1975) in some of the videos showing an interaction between two actors. In the social violation task, the participant is asked to understand the inadequacy of communicative acts with respect to the social context.
- **Production:** the subject must produce communicative acts that require a variety of levels of formality or informality.

Conversational scale

This scale evaluates the child's ability to entertain a conversation (with the experimenter), respecting topic, time, content and turn-taking. The examiner engages the participant in four short, free-flowing conversations on a variety of topics, such as leisure activities or summer holidays.

Evaluation of general intelligence

To avoid differences that may be due to overall general intelligence, we administered the Coloured Progressive Matrices test (CPM; Raven, 1947) to the two groups (CI and TH). Children had to solve 3 sets of 12 coloured puzzles (for a total of 36) by choosing the missing part among six alternatives. CPMs enable evaluation of intelligence based on problem-solving and logical thinking, without involving language ability.

4.2.3. Procedure

All children of both groups (experimental and control) were tested individually (only the examiner was in the same room with the child). HI children with CIs were tested individually in a room at the clinic, while TH children were tested individually in an empty classroom at school. Graduate research assistants in Psychology administered the tests (ABaCo and CPM) in two sessions, each lasting approximately one hour. The ABaCo was administered to both groups with the same identical procedure, i.e., participants were asked to watch the video recorded scenes with the examiner and then to answer some questions to show their understanding of the situation, or to complete an interaction. In case children did not understand or hear an instruction, or part of a video, the examiner repeated it or played the video again. The CPMs were presented in paper format, while the ABaCo test was presented on a laptop. None of the children received any help (i.e., use of loudspeakers). Reading and writing abilities were not involved in these tests. The order of the two tests was randomized.

4.2.4. Data Analysis

Statistical analyses were performed using R software and Psycho package (Core R Team, 2013).

Differences in communicative-pragmatic

To investigate differences in pragmatic performance between children with cochlear implants and peers with typical hearing development, we used a generalized linear model (GLM) with participants' scores on the ABAco as outcome, group of participant (two levels: children with cochlear implants, typically hearing peers), scale (five levels: linguistic, extralinguistic, paralinguistic, contextual, and conversational), and age group (three levels: 6;11 – 7;11; 8;0 – 8;11; 9;0 – 9; 11) as categorical predictors, level of intelligence as continuous covariate, and varying effects by participants and items. We tested the significance of each predictor and interaction term by performing an analysis of deviance (with type III Wald chi-square test) as implemented in the car package (Fox & Weisberg, 2011). Post-hoc pairwise comparison with Tukey correction for multiple comparisons was conducted using the lsmeans package (Lenth, 2016).

Role of the age of implantation and non-verbal intelligence

To analyse the role of the different factors explaining the pragmatic performance of children with cochlear implants on the ABAco Battery, we used a generalized linear model (GLM) with scores on the ABAco as outcome, separately evaluating the effect of relevant predictors - age, age of first and second implantation (in months), non-verbal intelligence (Raven matrices scores) for each ABAco scale (linguistic, extralinguistic, paralinguistic, contextual and conversational) - and varying the effects by participants and items. We included relevant predictors using a forward selection procedure, starting from a null model including just age (in months), and then checking at each step whether the addition of each predictor corresponded to a significant increase in goodness of fit using the likelihood ratio test and the Akaike Information Criteria (AIC). Continuous relevant predictors were scaled before being included in the model. The rationale of the procedure was to assess whether the

inclusion in the model of each relevant predictor, and of age of first and second implantation in particular, was able to improve the fit of the model and provide a unique contribution in explaining the poorer pragmatic performance of children with cochlear implants, having assessed the role of other relevant factors. This analysis was only performed on the group of children with cochlear implants.

4.3. Results

4.3.1. Multimodal pragmatic ability

The analysis revealed a significant effect of *Group* ($\chi^2(1, N = 44) = 11.31, p = .00$). As shown in Table 2.2, an examination of the main effects of *Group* revealed that overall, children with cochlear implants performed worse than typically hearing peers on the ABAco as a whole ($\beta = -.17, p = .00$). The analysis also showed a significant effect of *Scale* ($\chi^2(4, N = 44) = 129.52, p = .00$), with different levels of performance on the different scales of the ABAco. We also found a significant effect of *Age Group* ($\chi^2(2, N = 44) = 13.08, p = .00$), showing that overall performance on the ABAco improves with age in the 8 and 9-year-old groups. See Table 2.2.

The interaction between *Scale*Group* was also significant ($\chi^2(4, N = 44) = 11.77, p = .02$). Post-hoc pairwise comparisons with Tukey correction revealed that children with CIs (all age groups pooled together) performed worse than typically hearing peers on the paralinguistic ($p = .00$) and contextual scales ($p = .00$), while no significant differences were found on the linguistic, extralinguistic, or conversational scales (see Table 2.2).

2.2 PERFORMANCE OBTAINED BY CHILDREN WITH COCHLEAR IMPLANTS (CI) AND TYPICALLY HEARING PEERS (TH) ON THE DIFFERENT SCALES AND ON THE OVERALL ABaCo BATTERY IN THE DIFFERENT AGE GROUPS (6;11 – 7;11; 8;0 – 8;11; 9;0 – 9; 11). IN THE TABLE ARE REPORTED THE P-VALUE FOR POST-HOC TESTS WITH TUKEY CORRECTION FOR MULTIPLE COMPARISONS, AND THE EFFECT SIZE OF THE DIFFERENCES (HEDGES' G).

| Performance on the different scales and on the overall ABaCo battery | | | | |
|---|--------------------|--------------------|----------------|------------------|
| | CI (n = 22) | TH (n = 22) | p-value | Hedges' g |
| Overall ABaCo | .68 (.15) | .77 (.14) | NA | -.70 |
| Linguistic | .68 (.13) | .74 (.14) | .22 | -.50 |
| Extralinguistic | .65 (.14) | .68 (.14) | .53 | -.22 |
| Paralinguistic | .71 (.17) | .83 (.15) | < .001 | -.87 |
| Contextual | .67 (.24) | .83 (.20) | < .001 | -.82 |
| Conversational | .93 (.09) | .96 (.05) | .45 | -.57 |
| Performance on different scales and on the overall ABaCo battery in the different age groups | | | | |
| Age group (6;11 – 7;11) | | | | |
| | CI (n = 10) | TH (n = 9) | p | g |
| Linguistic | .58 (.17) | .69 (.18) | <.05 | -.64 |
| Extralinguistic | .61 (.12) | .68 (.16) | .23 | -.47 |
| Paralinguistic | .58 (.16) | .61 (.16) | .42 | -.18 |
| Contextual | .61 (.19) | .74 (.17) | <.05 | -.78 |
| Conversational | .53 (.28) | .73 (.28) | <.01 | -.77 |
| Overall ABaCo | .90 (.10) | .95 (.06) | .34 | -.94 |
| Age group (8;0 – 8;11) | | | | |
| | CI (n = 6) | TH (n = 6) | p | g |
| Linguistic | .72 (.06) | .80 (.06) | <.05 | -1.46 |
| Extralinguistic | .69 (.13) | .74 (.06) | .44 | -1.06 |
| Paralinguistic | .69 (.09) | .68 (.08) | .78 | .12 |
| Contextual | .76 (.11) | .88 (.09) | <.05 | -1.55 |
| Conversational | .74 (.17) | .88 (.10) | <.01 | -1.55 |
| Overall ABaCo | .92 (.08) | .96 (.07) | .28 | -.79 |
| Age group (9;0 – 9;11) | | | | |
| | CI (n = 6) | TH (n = 7) | p | g |
| Linguistic | .79 (.06) | .86 (.05) | .45 | -1.40 |
| Extralinguistic | .79 (.07) | .83 (.08) | .78 | -.43 |
| Paralinguistic | .74 (.07) | .78 (.09) | .93 | -.49 |
| Contextual | .82 (.08) | .91 (.07) | .23 | -1.38 |
| Conversational | .82 (.10) | .91 (.09) | .06 | -1.07 |
| Overall ABaCo | .98 (.04) | .96 (.06) | .51 | .41 |

The interaction between *Scale*Age group* was also significant ($\chi^2(8, N = 44) = 16.14, p = .04$). Looking at the differences in scores on the ABaCo scales for the different age groups (three levels: 6;11 – 7;11 vs 8;0 – 8;11 vs. 9;0 – 9; 11), post-hoc pairwise comparisons with Tukey correction revealed that children with a CI in the youngest age group (6;11 – 7;11) and intermediate age group (8;0 – 8;11) differed significantly from their hearing peers in terms of overall performance on the ABaCo ($p = .02; p = .04$) (see Figure 2.1).

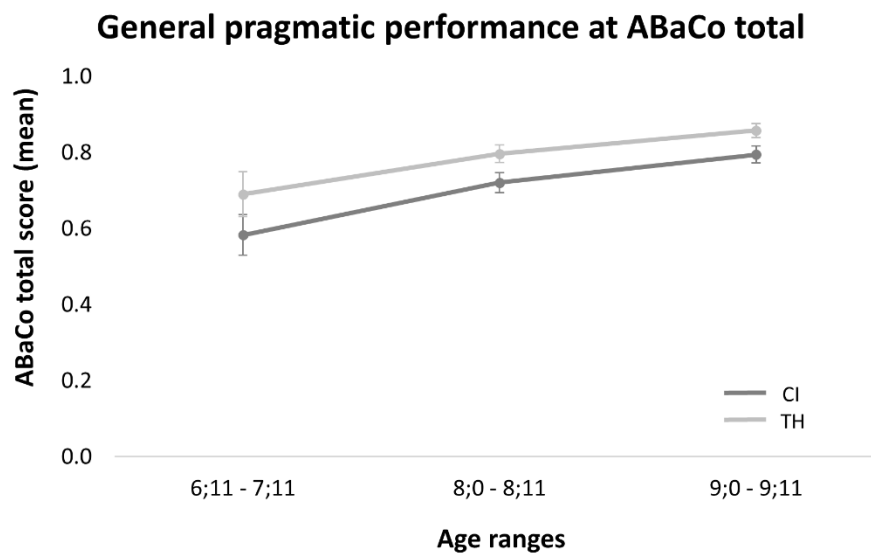


FIGURE 2.1 GENERAL COMMUNICATIVE-PRAGMATIC PERFORMANCE AT ABaCo. GENERAL COMMUNICATIVE-PRAGMATIC PERFORMANCE AT ABaCo ACROSS THE DIFFERENT AGE RANGES IN THE GROUP OF CHILDREN WITH COCHLEAR IMPLANTS (CI, DARK GREY) AND THE GROUP OF CHILDREN WITH TYPICAL HEARING ABILITY (TH, LIGHT GREY). ERROR BARS INDICATE STANDARD ERRORS.

Considering the different tasks in the different age groups, we found that children with a CI in the Age Group 6;11 – 7;11 differed from their hearing peers on the Contextual scale ($p = .0008$) and on the Paralinguistic scale ($p = .02$), children with a CI in the Age Group 8;0 - 8;11 differed from their hearing peers on the Contextual scale ($p = .00$) and on the Paralinguistic scale ($p = .02$), while no differences were observed between children with a CI and their hearing peers in the Age Group 9;0 – 9;11 (See Figure 2.2).

Participants performance on ABaCo scales

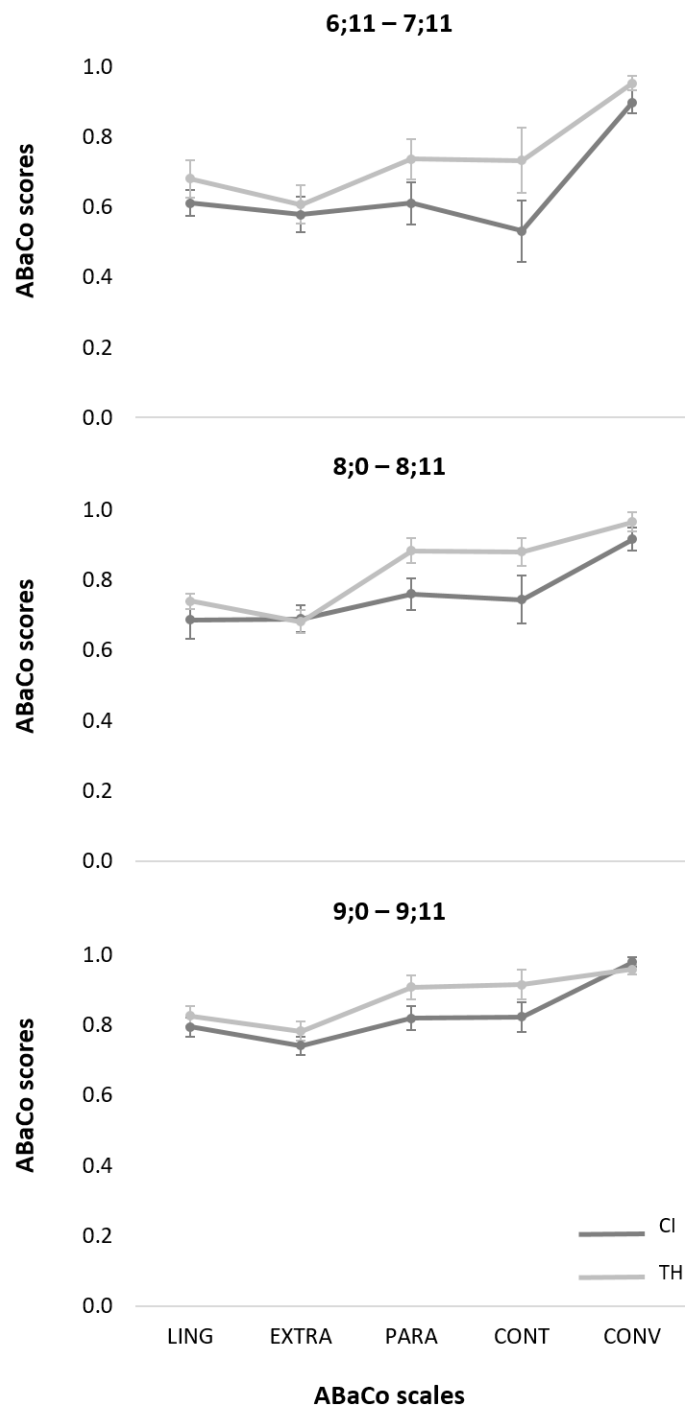


FIGURE 2.2 COMMUNICATIVE-PRAGMATIC PERFORMANCE AT ABaCo’s SCALES IN THE THREE DIFFERENT AGE-RANGES. COMMUNICATIVE-PRAGMATIC PERFORMANCE AT ABaCo’s SCALES IN THE THREE DIFFERENT AGE-RANGES IN THE GROUP OF CHILDREN WITH COCHLEAR IMPLANTS (CI, DARK GREY) AND THE GROUP OF CHILDREN WITH TYPICAL HEARING ABILITY (TH, LIGHT GREY). ABBREVIATIONS: LING = LINGUISTIC, EXTRA = EXTRALINGUISTIC, PARA = PARALINGUISTIC, CONT = CONTEXTUAL, CONV = CONVERSATIONAL. ERROR BARS INDICATE STANDARD ERRORS.

Finally, we also found an effect of the covariate CPM ($\chi^2(1, N = 44) = 9.39, p = .00$), indicating a role of non-verbal intelligence in pragmatic performance (both groups, i.e., CI and TH, pooled together). However, differences between children with a CI and TH peers still remained significant after controlling for the role of the covariate, i.e., level of intelligence.

4.3.2. Role of implantation and non-verbal intelligence

The analysis revealed a significant effect of age of first implantation ($\chi^2(5, N = 22) = 28.62, p = .00$) and of non-verbal intelligence ($\chi^2(5, N = 22) = 29.73, p = .00$) on overall pragmatic performance in children with cochlear implants. Considering separately the performance of children with CI at each single ABAco scale, we found that earlier age of implantation was associated with better performance on the contextual ($\beta = -.08, p = .00$) and paralinguistic scales ($\beta = -.06, p = .00$), and that higher non-verbal intelligence scores were associated with better performance on the contextual scale of the ABAco ($\beta = .12, p = .00$). Adding age of first implantation and non-verbal intelligence to the null model (including age only) improved the likelihood of the model and the AIC given our data. However, adding age of second implantation did not result in any further improvement in the likelihood of the model, and thus the selected model only included age, age of first implantation and non-verbal intelligence among the predictors.

4.4. Discussion

In the present research we provided a comprehensive assessment of communicative-pragmatic ability from a multimodal perspective in a group of children with cochlear implants and a group of typically hearing peers. We assessed the impact of bilateral cochlear implants on the development of a broad range of expressive modalities – linguistic,

extralinguistic, paralinguistic, contextual and conversational - in children with hearing loss, in different age groups - 6;11 – 7;11; 8;0 – 8;11; 9;0 – 9;11 - and we evaluated the relationship between age of cochlear implantation and pragmatic development.

While previous research has shown that children with HI have difficulties in communicative-pragmatic tasks (Jeanes et al., 2000; Paatsch & Toe, 2014; Tye-Murray, 2003), most of these studies focused on assessing conversational and linguistic skills, without considering non-verbal/extralinguistic and paralinguistic expressive means, thus not providing a complete assessment of communicative pragmatic ability. Further, only a handful of studies investigated communicative-pragmatic ability in children fitted with bilateral CIs at an early age, in order to determine the impact of early cochlear implantation on the development of pragmatic ability and the results of such studies are inconsistent and not conclusive (Guerzoni et al., 2016; Most et al., 2010; Nicastrì et al., 2014; Rinaldi et al., 2013; Socher et al., 2019).

The present study contributed to fill the existing gap in the current literature and to disentangle the role of the age of first and second cochlear implantation in the development of children's pragmatic ability. We found that children with CIs displayed different developmental trajectories compared to typically hearing peers. In particular, they showed poorer overall performance on the ABAco compared to typically hearing peers. This is in line with previous studies showing that children with hearing loss, even if fitted with a CI, continue to have some difficulties in social interactions and pragmatic abilities (Rinaldi et al., 2013). For example, Most et al. (2010) found that children of a similar age (6-9 years old) with HI, fitted with CIs or with hearing aids, were able to develop several pragmatic abilities at a level comparable to that of TH peers. However, they demonstrated inappropriate use of certain pragmatic behaviours, such as turn-taking and topic maintenance more frequently compared to typically hearing children. Nicastrì et al. (2014) pointed out that

children between 6 and 14 years of age with a unilateral cochlear implant performed less well in metaphor comprehension compared to TH peers, while no differences were found in tasks assessing discourse inferences. In line with this study, we found that even when children with CIs were able to display pragmatic behaviours, they performed less well than TH peers in different tasks included in the ABaCo, indicating a delayed development of pragmatic abilities.

We also wanted to assess the differences in pragmatic performance between children with a CI and typically hearing peers as a function of age group, that is to say, whether such differences are influenced by children's age. The results showed that differences in pragmatic ability are mediated by the children's age. Indeed, we found differences in overall pragmatic ability assessed with the ABaCo between children with CI and typically hearing peers in the two younger age groups of children, 7- and 8- years, but not in the older one of 9-years. This datum indicates that while the differences are greater in the younger groups (with medium effect size) and suggest a delay in the development of pragmatic skills, they tend to narrow and be less evident in children with CIs in the oldest age group, whose pragmatic ability was comparable to that of typically hearing peers. We explain (in line with Toe et al. 2007) this developmental trend as due to different factors such as the longer exposure to auditory stimuli combined with the higher level of engagement in very structured and well-contextualized social interactions with peers and teachers at school experienced by older children.

By focusing on the different scales of the ABaCo, we found significant differences in the performance of children with CIs compared to TH peers (all age groups pooled together) on two scales of the ABaCo, i.e., the Paralinguistic and Contextual scales. The Paralinguistic scale assesses the comprehension and production of those communicative aspects that complement the interaction, such as tone of the voice and prosody, and that

could convey the speaker's real communicative intentions. Our outcome is in line with previous research (Le Maner-Idrissi et al., 2020) assessing emotional speech comprehension in children with CIs. These children have serious difficulties in the processing of acoustic inputs that could lead to the construction of atypical phonological representations, as well as to difficulties in handling those linguistic elements which rely more on acoustic processing, such as rhythm or accents (e.g., unstressed bound, free morpheme, see Hammer et al., 2010). Impaired acoustic processing can negatively reflect in the recognition and production of prosodic (verbal and non-verbal) aspects of conversation, such as the use of the tone of voice to convey emotional aspects, mark relevant information or express different basic speech acts (e.g., questions or orders), which we found to be impaired in the group of children with CIs. The Contextual scale evaluates the adequacy of a communicative act with respect to the norms of discourse (i.e., Grice's maxims) and social norms of communication. Previous studies have shown that children with CIs are less exposed to conversation than TH peers (see Paatsch et al., 2017). Indeed, analyses of parent-child conversations have revealed that conversations between children with CIs and their parents tend to be more controlled. Parents of children with CIs tend to allow their children less time to talk, and such children rarely start a conversation of their own accord, and this reduces the opportunities for them to be engaged in conversations with peers or other adults (Rinaldi et al., 2013). Thus, the reduced exposure to social interactions may partially explain the delayed ability of these children to recognize social norms of communication, which are generally learned during communicative exchanges.

Consistently with Ambrose (2016), we found no differences in performance on the extralinguistic scale between children with CIs and TH peers, indicating that children with CIs are able to develop abilities that do not require the processing of a linguistic input in a normal range. This finding points to the importance of considering alternative

communicative modalities in training aimed at promoting the development of linguistic-pragmatic skills. We also found no significant differences in performance on the linguistic or conversational scales, in contrast with previous studies that reported differences between children with HI and TH peers in these abilities (Church et al., 2017; Tye-Murray, 2003).

Overall, these results suggest that the delay in the development of pragmatic ability is not uniform across the different scales of the ABaCo, with children with CIs showing a level of development comparable to that of TH peers in some pragmatic skills but not in others; furthermore, these differences are not uniform across the different age groups, with older children's performance on the ABaCo not differing from that of TH peers. These results suggest the need for a comprehensive and articulated assessment of pragmatic abilities when dealing with children with HI, in order to detect strengths and weaknesses in the development of pragmatic skills, and indicate the importance of tracking developmental trajectories by assessing pragmatic ability at different ages.

We also found a significant role of the covariate non-verbal intelligence on pragmatic performance. This is in line with previous studies showing that non-verbal intelligence is generally associated with language acquisition (Watson et al., 1982). However, our primary aim was to assess whether differences in pragmatic-performance between children with CIs and TH peers might be due to differences in non-verbal intelligence. In this respect, we found that differences in pragmatic performance are still present even after controlling for the role of non-verbal intelligence. This finding is in line with both developmental studies and studies on different clinical populations which demonstrated the specificity of pragmatic impairment over and beyond the role of non-verbal intelligence and other cognitive functions (Bosco & Gabbatore, 2017a; Parola et al., 2020).

The second aim of our investigation was to evaluate the effect of age at implantation on pragmatic development. To this end, we included children who had undergone bilateral

cochlear implantation at different ages and we assessed how the age of first and second implantation was able to predict the development of pragmatic abilities during the early school years. Our result indicated that age of first implantation is the best predictor of pragmatic performance in children with CIs, even after controlling for the other confounding factors, i.e., level of intelligence and children's age, while adding age of second implantation did not improve the fit of the model any further. This result showed that children with CIs who received their implant at an earlier age performed better on the ABAco. More specifically, we found the age of first implantation is related to the pragmatic performance on the two ABAco scales for which we found significant differences between children with CIs and TH peers, i.e., the paralinguistic and contextual scales. This result confirmed that the earlier the children were fitted with the implant, the lesser the delay they experienced in these pragmatic skills. This result is in line with previous evidence indicating that age at implantation is a key factor in influencing the development of communicative ability (Guerzoni et al., 2016; Tye-Murray, 2003). For example, Nicastrì et al. (2014) showed, in a group of children with CIs comparable to those of our study in terms of age and age of implantation, that age at implantation was significantly correlated with the development of different pragmatic skills, such as comprehension of metaphors and implicit meaning. However, other studies have reported different results (Socher et al., 2019). For example, Rinaldi et al. (2013) compared children who received the CI by 12 months of age with those implanted during the second year of life, and did not find any significant difference in vocabulary size or early grammar skills between the two groups. Inscoe et al. (2009) found no role for age at implantation in expressive spoken language skills. However, it should be noted that there are important differences across the different studies reported above in terms of mean age of implantation, implantation modality (unilateral vs. bilateral), rehabilitation therapy (therapy vs. no therapy), the linguistic/pragmatic skills assessed, and children's

mean age, and it is thus difficult to make direct comparisons across these studies. Even if the evidence supporting the importance of early implantation is far from conclusive, it does indicate that age of first implantation is a crucial factor in the development of pragmatic abilities, and that early implantation can promote typical communicative-pragmatic development. Future studies should consider this aspect and try to replicate these results. Furthermore, it should be noted that along with the age of implantation, speech therapy also provides crucial help in the development of communicative abilities, especially auditory-verbal therapy which emphasizes linguistic abilities in both comprehension and production (Hogan et al., 2008).

A limitation of the present study was the small number of participants. This reduced the statistical power of the analysis and the possibility of detecting significant differences, especially for comparisons across the three different age ranges. Furthermore, we did not collect any measurements of the children's vocabulary skills – such evaluations might have been useful for an overall and complete assessment of their communicative abilities, and for understanding how these abilities develop over time. Further, the adoption of a longitudinal design would have allowed us to test more specific hypotheses on changes in pragmatic skills over time. Finally, an assessment of other social skills, such as Theory of Mind, i.e., the ability to ascribe thoughts, beliefs and desires to one's self and to others (Premack & Woodruff, 1978), would have allowed us to conduct a more detailed analysis of the factors responsible for the delayed acquisition of communicative-pragmatic skills in children with CIs. These aspects should thus be addressed in future studies, and the present results need to be replicated with a larger sample and adopting a more principled design able to track the development of pragmatic abilities over different time points.

To conclude, our results show that bilateral CIs are helpful in reducing the differences in pragmatic abilities between children with HI and TH peers. More in detail,

children with CIs produced responses similar to those of TH peers on the linguistic, extralinguistic and conversational scales of the ABaCo, even if some difficulties still remain in specific pragmatic aspects as the ability to handle paralinguistic cues and the sensitivity to social contextual ones. The delays in the development of pragmatic skills are mediated by age at implantation, with children fitted with a CI at an earlier age achieving a level of pragmatic performance similar to that of TH peers. Our findings pinpoint the importance of a complete and principled pragmatic assessment in children with CI, in order to identify their strengths and weakness, and the role of an early cochlear implantation, which, combined with a speech therapy, contributes to foster a typical development of their communicative-pragmatic ability.

PART B

Neurodevelopmental disorders:

Autism spectrum disorder

5. Introduction

Autism spectrum disorder (ASD) is a complex neurodevelopmental condition that affects social communication and social interaction. It is characterized by repetitive behaviours and restricted interests or activities that can persist throughout life (DSM-5; APA, 2013), with an impact on social, academic and professional/occupational life. According to the WHO meeting report entitled “Autism spectrum disorders & other developmental disorders” the estimated global incidence of ASD is 1/160 persons (WHO, 2013) with a prevalence in males (3:1) (Loomes et al., 2017)¹¹. The label “spectrum” includes pervasive developmental disorder (PDD), i.e., autistic disorder, Asperger’s disorder, childhood disintegrative disorder, and pervasive developmental disorder not otherwise specified (PDD-NOS) (Hodges et al., 2020). The DSM-5 guidelines suggest how to categorize the level of impairment (APA, 2013):

- Level 1 “*Requiring support*”: individuals show decreased interest and difficulty in initiating social interactions and functional difficulties caused by a lack of flexibility in behaviours;
- Level 2 “*Requiring substantial support*”: there is significant difficulty in communicative abilities even when receiving some help and support and an impossibility to face and cope with change;
- Level 3 “*Requiring very substantial support*”: there is severe impairment of general functioning caused by a very limited use of social interactions and communication, and extreme difficulty in adjusting behaviours and focusing attention.

¹¹ It is worth noting that the study by Loomes and colleagues (2017) used data collected since the introduction of the DSM-IV. However, some studies suggested that the introduction of the new definition present in the DSM-V could decrease ASD incidence due to the change in diagnostic criteria (Kulage et al., 2014; Maenner et al., 2014; McPartland et al., 2012).

Deficits in theory of mind (ToM; Baron-Cohen et al., 1985; Chung et al., 2014; Hoogenhout & Malcolm-Smith, 2017; Jones et al., 2018) and executive functions (EFs; Demetriou et al., 2018; Demetriou et al., 2019; Panerai et al., 2016; Robinson et al., 2009) are also associated with ASD.

ToM refers to the ability to attribute to oneself and to others mental states, emotions, beliefs, and desires (Premack & Woodruff, 1978) in order to make behavioural predictions. A variety of theories to describe ToM have been advanced (Baron-Cohen et al., 1985; Leslie, 1987, 1994). Furthermore, a deficit in ToM development has also been theorised to be the origin of ASD symptoms, especially social communication (Baron-Cohen et al., 1985).

Executive functions are a set of cognitive abilities that control behaviours in everyday activities and that allow attainment of goals. Among the accounts to explain executive function (Diamond, 2013; García-Madruga et al., 2016; Miller & Cohen, 2001), Miyake et al. (2000) identified three main cognitive components: *shifting*, *updating*, and *inhibition*. Shifting refers to the ability to shift attention to different tasks, updating refers to the ability to monitor and manipulate mental representations, and inhibition is the ability to repress predominant and non-significant stimuli (Miyake et al., 2000).

Connected to executive functions is weak central coherence (WCC), which has also been proposed to explain ASD symptoms (Frith, 1989; Happé & Frith, 2006). WCC refers to the tendency to process information by focusing on details rather than on the global context, which appears to characterise ASD behaviour. Although studies have shown that WCC and executive functions are independent of one another (Booth et al., 2003; Teunisse et al., 2001), other research has raised doubts about this lack of association (Motttron et al., 1999; Pellicano, 2010; Vanegas & Davidson, 2015).

The association between ToM and EFs and their role in ASD symptomatology are not completely understood (Joseph & Tager-Flusberg, 2004). Jones and colleagues (2018) investigated the relationship between ToM and EFs in adolescents with ASD and found an association between ToM and social communication deficits and repetitive behaviours. In contrast, EFs that correlated with ToM did not have a direct relation with ASD symptoms. In her study, Pellicano (2007) reported that children with ASD may present intact EFs and a significant impairment in ToM, suggesting that EFs do not rely on ToM ability. Nevertheless, in a study by Ozonoff and colleagues (1991), EFs deficits appeared to be more pervasive in ASD than a ToM deficit. Furthermore, interventions for treating EFs have been found to improve ToM, while the opposite effect (i.e., ToM training to enhance EFs) has not been observed (Fisher & Happé, 2005). In brief, debate surrounds the nature of the relationship between these two cognitive domains. Further study is needed in order to disentangle this interconnection (Wade et al., 2018).

Though the causes of ASD are not well defined, several risk factors have been identified: genetic factors (Rylaarsdam & Guemez-Gamboa, 2019), advanced parental age, maternal history of autoimmune disease, maternal infection during pregnancy, premature birth, issues related to childbirth (see also Hodges et al., 2020). Since causal determinations are not possible at the moment, scientific and clinical communities have focused their attention on early diagnosis.

The first symptoms of ASD appear in the early stages of development; nonetheless, the diagnosis might be difficult to make because some impairments emerge when the child starts to engage in social contexts and situations. Furthermore, some features characterizing the disorder may be difficult to distinguish from those of other disorders (Baio et al., 2018). Major attention has been directed to interventions and rehabilitation to reduce the burden of this disorder.

5.1. Communication in adolescents with autism spectrum disorder

Individuals with ASD show a deficit in social communication and a wide variety of other difficulties. First, a general delay in language development has been observed in toddlers with ASD (Özyurt & Eliküçük, 2018; Wetherby et al., 2004) characterized by less frequent bubbling (Patten et al., 2014) and greater production of atypical non-speech vocalizations (i.e., screams or crying without recognizable consonants) than typically developed children (Schoen et al., 2011). Anomalies in the development of syntax are noted in the incorrect interpretation of reflexive pronouns (Perovic et al., 2013), the use of relative clauses (Durrleman et al., 2015), and *wh*-questions (Durrleman et al., 2016; Goodwin et al., 2012). Children with ASD who are not affected by syntactic impairment manage to develop a rich lexicon (McGregor et al., 2012). But even when syntactic ability is spared, children with ASD still present difficulties with semantics and especially with pragmatics (Ambridge et al., 2021; Sukenik & Tuller, 2021).

Impairment of communicative competence is well documented in the literature (Baixauli-Fortea et al., 2019; Martzoukou et al., 2017; Volden, 2017). Individuals with ASD have trouble dealing with various aspects of the pragmatic domain, such as humour, sarcasm, and irony (Emerich et al., 2003; Persicke et al., 2013; Saban-Bezalel et al., 2019; Wu et al., 2014; Zalla et al., 2014), figurative language (Kalandadze et al., 2018), e.g., metaphors¹² (Kalandadze et al., 2019; Rundblad & Annaz, 2010), and indirect speech (Marocchini et al., 2021). Also, ASD affects other expressive modalities besides the linguistic channel. Empirical evidence suggests that both extralinguistic (Colgan et al., 2006; Morett et al., 2016; Silverman et al., 2010) and paralinguistic abilities (McCann & Peppé, 2003; Shriberg et al., 2001) are impaired in ASD (Angeleri et al., 2016).

¹² It is worth noting that some studies by Kasirer and Mashal (2014, 2016) reported that the deficit in metaphor comprehension and production is related to conventional metaphors but not necessary to novel metaphors.

Another component of communicative ability that is impaired in ASD is the capacity to engage in conversation and to produce discourse and narrative. Individuals with ASD show deficits in the ability to take active part in a conversation and follow conversational and social norms, adhere to discourse topic, and use turn takings appropriately (Angeleri et al., 2016).

Previous studies have reported poor narrative ability in verbally fluent children with ASD (Carlsson et al., 2020; for a meta-analysis, see Baixauli et al., 2016). For instance, at the microlinguistic level, children with ASD produce more grammatical errors and less complete sentences, whereas at the macrolinguistic level, they may fail to use linking devices correctly (e.g., referential pronouns and conjunctions) in organising story content (Makinen et al., 2014; Westerveld & Roberts, 2017).

There is some evidence for the persistence of narrative deficits in young-adults (Rollins, 2014) and adults with ASD (Colle et al., 2008; Geelhand et al., 2020); however, study findings about the narrative profile of individuals with ASD during adolescence are inconclusive. Studies to date that have investigated narrative production abilities in both children and adolescents with ASD report that they make considerably more micro- (e.g., reduced syntactic complexity) and macrolinguistic (e.g., irrelevant comments and ambiguous references) errors than their typically developing peers (Losh & Capps, 2003; Marini et al., 2020). While such difficulties may decrease with age (Iandolo et al., 2020; Norbury et al., 2014), a meta-analysis by Baixauli et al. (2016) found that the narrative performance of children and adolescents (up to age 15) with ASD was not predicted by age, suggesting that narrative difficulties in ASD are likely to persist from childhood through adolescence.

Moreover, studies investigating the narrative profile of teenagers with ASD have reported that syntactic oversimplification persists through adolescence and that the ability to

express temporal and causal connections between events is low in adolescents with ASD (Broc et al., 2021; King & Palikara, 2018). Difficulty in building textual cohesion and managing coherent narrative discourse (Canfield et al., 2016; King & Palikara, 2018) make it harder to understand the stories of adolescents with ASD compared to those produced by their typically developing peers (de Marchena & Eigsti, 2016).

The interplay between communicative abilities and other cognitive components (e.g., ToM and EFs) has been extensively investigated, although the nature of these relationships remains unclear. Effective and successful communication relies on the ability to recognise the attribution of intentionality, which is associated with ToM. A not-yet-fully developed ToM ability could be related to impaired narrative production in individuals with ASD. The ToM deficit may hinder them from understanding the listener's previous knowledge and from providing sufficient information for the interlocutor to completely understand the story. Also, this type of deficit can preclude individuals with ASD from understanding the motivation and the mental and emotional states that the characters of a figure/story might have (Tager-Flusberg & Sullivan, 1995). Studies report that children and young adolescents with ASD experience more difficulty in identifying the mental states of story characters than healthy subjects and that their poorer performance is related to ToM measures (Capps et al., 2000). Evidence for the relationship between ToM and pragmatics and narrative, is contradictory, however. Sperber & Wilson (2002) define pragmatics as a subcomponent of ToM: inferential processes can be viewed as a series of metapsychological processes of attribution of communicative beliefs and intentions (Sperber & Wilson, 2002). Differently, other theoretical approaches state that the role of ToM cannot be attributed only to the processes of recognizing the intention of others. While it is clear that the ability to attribute mental states to others is important for the performance of pragmatic tasks, communicative-pragmatic ability does not seem to be entirely dependent on and determined by ToM alone

(Bosco et al., 2018; Bosco, Tirassa, et al., 2018; Cardillo et al., 2021; Domaneschi & Bambini, 2020; Matthews et al., 2018). Consistent with the last cited studies, recent findings showed no improvement in the pragmatic performance of persons with ASD after treating ToM (Marraffa & Araba, 2016). The lack of improvement underscores the importance of focusing not only on the similarities but also the differences between the two abilities since they have different implications for the clinical domain.

Communicative ability is also associated with EFs: to achieve successful communication, individuals must keep and manipulate mental representations and information by using their working memory skills, plan and organize the discourse correctly, adapt to the evolution and the direction of the conversation by shifting skills and flexibility, repress their own perspective and consider that of others. When producing a story, multiple cognitive skills help to organize it, inhibit the irrelevant details and select the salient ones, and switch from the description of one event to another by connecting them. Kuijper and colleagues (2015) found that working memory and inhibition were predictive of appropriate referent reintroduction in narrative production tasks; Ketelaars and colleagues (2012) showed that executive function is predictive for narrative productivity, when controlling for language ability, in children with pragmatic language impairment, while no specific link between such skills was found in children with typical development. Analogously, Blom and Boerma (2016) measured narrative comprehension and production in children with developmental language disorder and found an association between working memory and narrative comprehension and production, while working memory in typically developing children was associated with comprehension only. Losh and Gordon (2014) evaluated narrative abilities in children and young adolescents with ASD and found that difficulty on a narrative recall task may be related to reduced attention. Furthermore, disorders that manifest with pragmatic impairment are accompanied by deficits in executive functions,

which suggests their involvement in pragmatic ability (Bambini et al., 2021; Bosco et al., 2018; Bosco, Parola, Sacco, et al., 2017; Champagne-Lavau & Stip, 2010). Finally, narrative appears to be connected also to the weak central coherence: individuals with ASD are noted to show a tendency to process details rather than integrate them in a global meaning. They may find it difficult to incorporate the context when inferring a meaning (Kenan et al., 2019; Vermeulen, 2015), which can lead them to focus more on details rather than on the story plot.

Overall, the current literature suggests that cognitive functions correlate with (some aspects of) pragmatic and narrative ability, while these address specific aspects and are not merely the sum of different cognitive abilities (Bosco, Tirassa, et al., 2018; Domaneschi & Bambini, 2020).

Little attention has been directed to specific interventions on the narrative abilities in adolescents with ASD (Parsons et al., 2017). General communicative treatment for adolescents with ASD employs various different strategies, including peer-mediated conversational treatment (Axe, 2018; Bambara et al., 2016; Thomas & Bambara, 2020), social skills group training (Choque Olsson et al., 2017; Dekker et al., 2019; Matthews et al., 2020), and treatment focused on communicative-pragmatic ability (Gabbatore et al., 2021). Only one study to date has described treatment-related improvement of narrative production in adolescents and young adults with ASD (McCabe et al., 2017). The study participants received a parental-mediated intervention to enhance the production of their personal narratives of everyday situations (e.g., visiting the hospital, getting lost, etc.). In their study, McCabe and colleagues trained the participants' parents and provided them with specific recommendations to promote narrative production in daily communicative interactions with their children (see also Peterson et al., 1999 for a similar procedure). Samples of narrative discourse produced by the participants before and after the treatment

were assessed by means of a quantitative (e.g., assessment of story grammar complexity, topic maintenance, occurrence of off-topic comments) and a qualitative approach (i.e., quality of the stories as perceived by their parents). Overall, an improvement in the narrative skills was observed. Though the outcome was positive, the treatment did not directly involve the participants with ASD but rather only their caregivers.

6. Study three: Narrative ability in adolescents with autism spectrum disorder receiving the Cognitive-Pragmatic Treatment¹³

6.1. Aims and hypothesis

Aim of this study is to determine whether a communicative-pragmatic rehabilitation program, i.e., the Cognitive Pragmatic Treatment (CPT; Gabbatore et al., 2015) adapted for adolescents (A-CPT; Gabbatore et al., 2021), is effective in enhancing the narrative skills in a cohort of verbally fluent adolescents with ASD. The aim of an A-CPT program is to improve communicative-pragmatic skills (e.g., conversational abilities, social awareness, production and comprehension of literal and non-literal communicative acts, such as indirect speech acts and irony) via a variety of expressive means (linguistic, extra-linguistic, e.g., gestures, and paralinguistic, e.g., prosody). The original version of the CPT has proven effective in increasing the narrative ability of persons with traumatic brain injury (Bosco et al., 2018; Parola et al., 2019; Sacco et al., 2016). Recently, A-CPT has been shown effective in improving pragmatic abilities in a cohort of verbally fluent adolescents with ASD by a pre- post- specific evaluation with the equivalent form of the Assessment Battery for Communication (Bosco et al., 2012) - in both comprehension and production (Gabbatore et al., 2021). That said, data are missing on the potential positive effect of A-CPT on the narrative skills of individuals with ASD.

¹³ Portions of text and data of this chapter are currently submitted and under review as “Hilviu, D., Frau, F., Bosco, F. M., Marini, A., Gabbatore, A. Can narrative skills improve in autism spectrum disorder? A preliminary study with verbally fluent adolescents receiving the Cognitive Pragmatic Treatment” to the *Journal of Psycholinguistic Research*.

To fill this gap, we assessed the narrative skills of a cohort of adolescents with ASD pre- and post-training using a multilevel approach to discourse analysis (Marini et al., 2011). This approach was effective in capturing discourse errors in cohorts of children and adults with communicative impairment (Marini et al., 2007, 2008, 2010, 2020). We hypothesized that a substantial improvement in narrative skills at the micro- and the macrolinguistic levels would be noted in the study sample after participation in the A-CPT program.

In addition to the narratives, we assessed cognitive and ToM functioning. A recent narrative review (Matthews et al., 2018) found that, in typical and atypical development, the relationship between cognitive variables is not fully clear yet. Given the complex, and not yet completely clear relationship between these variables, we also assessed a pool of cognitive and ToM functions before and after training in order to verify that the effect of the training was specific for the target variable of the study, i.e., narrative ability.

Cognitive assessment was performed using a selection of tasks of the Neuropsychological Evaluation Battery (BVN 12-18; Gugliotta et al., 2009). The tasks evaluate memory, attention, and executive functions (i.e., *shifting, inhibition, planning*). ToM was assessed using tasks tapping first-order ToM and the ability to recognise emotions. As the above-mentioned cognitive abilities are not a target of A-CPT, the aim of these assessments was to determine whether potential improvement could be observed after specific training for pragmatic ability was delivered, which was the program target variable. Since we used the cognitive and ToM assessments as control measures, we did not expect any specific improvement allowing us to rule out a determinant role of ToM or cognitive functions in explaining the expected narrative improvement.

6.2. Material and Methods

6.2.1. Participants

Eighteen (17 males and 1 female) adolescents with a diagnosis of autism spectrum disorder made by an expert psychiatrist and based on DSM-IV criteria (ASD; American Psychiatric Association, 1994), were enrolled in the study through the collaboration between the research group and two rehabilitation centres in Piedmont (Italy) area, namely Gruppo Asperger Onlus (Turin) and Centro di Riabilitazione Ferrero (Alba, Cuneo). One participant did not complete the rehabilitative training because of personal commitments. Another participant was excluded from analysis due to a serious form of stuttering that made it extremely difficult to understand the recordings and encode the narratives. The final sample included 16 participants (15 males and 1 female; age range 12-18 years, mean 13.94 ± 1.98) with 6 to 13 years of schooling (mean $8.75 \text{ years} \pm 2.02$). All participants were native Italian speakers. They were initially enrolled based on their IQ (cut off ≥ 80) as reported in their clinical records. Nevertheless, they were retested with the Italian version of Raven's Standard Progressive Matrices (Raven, 1938) with reference to the standardized norms for adolescents (Picone et al., 2017). The mean IQ for the sample was 94.56 ± 14.30 . Individuals were excluded if they were attending an Applied Behaviour Analysis rehabilitation program or other programs targeting communicative abilities at the time of the present study. Inclusion criteria were adequate linguistic abilities, which were further assessed with the Token test of the Neuropsychological Evaluation Battery (BVN 12-18; Gugliotta et al., 2009), a subtest for linguistic comprehension.

The participants' families agreed to take part in the training program after they attended a presentation of the research project held at the rehabilitation centre and involving research group members, professionals working at the centre, adolescents and their families. Prior to data collection, the participants and their caregivers gave written, informed consent

to participate in the training program and permit videorecording of the sessions. The participants and their families were provided detailed information about the nature and aims of the study in compliance with the ethical code of the Italian Association for Psychology (AIP) and in accordance with the tenets of the Declaration of Helsinki. The participants and their families were also informed that participation was voluntary, that they could refuse to participate and withdraw from the study at any time, and that data confidentiality would be assured according to current data protection norms and legislation. The project was approved by the Bio-Ethical Committee of the University of Turin, protocol n. 144,890.

The training was provided in small groups of 4 to 5 participants, each undergoing through the same number of sessions and the same type of training activities. The average attendance rate was 94.27 %. The trainers were graduate students on a master's program in psychology; they had received training in the structure and the procedures of the A-CPT program, as well as in the administration of assessment tools for evaluating narrative, cognitive and ToM abilities. Assessment and rehabilitation were supervised by a team of experts in pragmatic and cognitive impairment, psychologists, and members of the research team who had developed the original CPT.

6.2.2. The rehabilitative program: CPT for adolescents

We used the version adapted for adolescents (A-CPT; Gabbatore et al., 2021) of the Cognitive Pragmatic Treatment (CPT; Gabbatore et al., 2015), a manualized treatment that has proven effective in enhancing communicative-pragmatic skills in a cohort of verbally fluent adolescents with ASD, aged 12-18. The A-CPT is a group training program theoretically grounded on Cognitive Pragmatics (Airenti et al., 1993a, 1993b; Bara, 2010), a theory on the cognitive and inferential processes underlying human communication. A-CPT focuses on communicative-pragmatic abilities, and specifically on a range of skills that

allow individuals to communicate efficiently and effectively: linguistic, extralinguistic, and paralinguistic abilities; social appropriateness; awareness; conversational and narrative skills; social and planning abilities. A-CPT includes activities designed to improve participants' communicative efficiency in both comprehension and production (Table 3.1). Each session has its focus on a specific aspect of communication and provides participants with an ecological setting where they can practice their pragmatic abilities while simulating daily routine activities. The aim of A-CPT activities is to help participants improve their inferential skills, i.e., their ability to fill the gap between literal and intended meanings (e.g., irony and figurative language). Another aim is to improve the ability to maintain attention through the efficient use of expressive modalities, i.e., language, gestures, facial expressions, prosody, tone and rhythm of the voice. Such an ability can better emphasize intended meaning and facilitate the understanding of pragmatic phenomena, e.g., in the use of irony, as an identical literal utterance may convey different meanings depending on situational cues (Bosco et al., 2017).

The structure of each session remained constant throughout the training program regardless of the specific topic of the session:

- *Introduction and overview.* Introduction to the current session content, with particular attention to the connection between the current communicative topic and participants' daily life episodes.

- *Comprehension activities.* These were mostly video clips illustrating brief communicative interactions created ad hoc for the A-CPT program. Participants were asked to observe two actors interacting in a specific communication modality presented during the session (i.e., linguistic modality in linguistic sessions or gestural modality in extralinguistic sessions and so forth). At the end of each video clip, the participants were invited to comment on the interactions they observed, with the aim to stimulate their comprehension of the

communicative situations portrayed during the video. Discussion with other group members served to improve discourse coherence and enhance compensatory communication strategies. Self-monitoring and feedback were provided by the therapist and the other group members during the training sessions to guide and support comprehension.

- *Production activities.* These were chiefly role-play activities (i.e., interactive scenarios of everyday situations), in which the participants held in-pairs communicative interactions to improve and strengthen their ability to use contextual elements. The role-play activities also provided the participants with communication strategies and feedback from the therapist and the other group members within a safe group-training setting. The A-CPT program included specific activities involving paralinguistic (e.g., recognition and production of facial expression, exercises for modulating the tone of voice), narrative (picture description and famous movie plots), and planning abilities (planning of activities and tasks to be performed within a given amount of time).

- *Conclusion and homework.* Assigned at the end of each session, the homework consisted of tasks for practice at home of the communication strategies that had been illustrated during the training session. The activities provided the participants with an opportunity to practice and improve their communicative skills acquired during the A-CPT sessions.

Overall, the A-CPT includes a total of 15 sessions: one session per week, each lasting approximately 90 minutes, including a 10-minute break (Table 3.1). The original version of the CPT (Gabbatore et al., 2015) consists of two sessions per week (12 weeks) for a total of 24 sessions; each session lasts approximately 90 minutes and includes a 10-minute break. See Bosco et al., 2016, 2018; Gabbatore et al., 2017; Sacco et al., 2016 for a more detailed description of the structure of the CPT and the content of rehabilitative sessions.

The length and the content of the original version of the CPT (Gabbatore et al., 2015) was adapted in order to make it more suitable for adolescents. The adapted version has been used in a previous study involving adolescents with ASD, in which it was shown effective in improving pragmatic skills in comprehension and production (Gabbatore et al., 2021).

TABLE 3.1 OUTLINE OF THE ADOLESCENTS ADAPTED VERSION OF THE COGNITIVE PRAGMATIC TREATMENT (A-CPT)

| TRAINING SESSION | DESIGNED ACTIVITIES |
|---|--|
| 1 INTRODUCTION AND OVERALL COMMUNICATIVE ABILITY | Introduction to the aims and structure of the CPT program; setting-up of the working group by a self-introduction of each participant, including the description of any perceived difficulty in daily living communication. Overview of the communicative-pragmatic ability, via video clips and role-playing tasks, based on daily living situations and depicting all the communicative expressing means. |
| 2 LINGUISTIC ABILITY | Video clips and role-playing based on the linguistic expressive modality. |
| 3 EXTRALINGUISTIC ABILITY | Video clips and role-playing based on the gestural modality. |
| 4-5 PARALINGUISTIC ABILITY | Video clips, facial expression recognition, tone of voice tasks, role-playing; |
| 6-7 SOCIAL APPROPRIATENESS | Video clips and role-playing focused on social appropriateness and communicative adequacy in different contexts. |
| 8 CONVERSATIONAL ABILITY | Video clips, role-playing and exercises focused on the use of conversational rules (i.e., turn-taking topic management). |
| 9 PHONE CONVERSATION | Audio clips and role-playing focused on telephone conversational rules (i.e., voice only, no paralinguistic and gestural clues, available in live interactions). |
| 10-11 SOCIAL ABILITY | Video clips and role-playing focused on the ability to formulate meta-representations with respect to one's own and others' mental states. |
| 12 NARRATIVE ABILITY AND PLANNING | Picture-description task, aimed at eliciting story-telling by providing an adequate amount and type of information. |
| 13-14 OVERALL COMMUNICATIVE ABILITY | Video clips and role-playing focused on the overall pragmatic effectiveness, expressed through all the modalities constituting communicative competence. |
| 15 CONCLUSION, AWARENESS AND FEEDBACK | Conclusions and feedback about the progresses observed along the CPT, i.e., video recording of the salient moments along the sessions where the improvements could be detected were shown to each participant during the group session |

6.2.3. Assessment measures

Narrative assessment

Narrative abilities were assessed at T0 (pre-training) and T1 (post-training), i.e., within one week after the end of the training. Discourse samples were elicited using four picture stimuli: two single-picture scenes entitled the Picnic taken from the Western Aphasia Battery (Kertesz, 1982) and the Cookie Theft by Goodglass and Kaplan (1972), and two picture sequences entitled the Flower Pot by Huber and Gleber (1982) and the Quarrel by Nicholas and Brookshire (1993). Each participant was assessed individually in a quiet room at the rehabilitative centre. Administration of the stimuli, transcription of the speech samples, and multilevel discourse analysis were performed following the criteria detailed in Marini et al. (2011). Pictures were administered in the same order to all participants by means of a laptop with the display facing the participant to prevent memory limitations and referent sharing. The participants had to describe the situation depicted in the pictures without using ambiguous words (e.g., *here*, *there*, etc.) as the task administrators stated they were unfamiliar with the stimuli. The narratives were audio-recorded. The speech samples were later transcribed verbatim by one transcriber, with the inclusion of phonological fillers, pauses, false starts, and extraneous utterances. The transcripts were analysed by the same coder. The duration (in seconds) of each sample was calculated, as well as the total number of *units* and *words*. The term *units* defines the verbalizations produced by a speaker, including well-formed words, non-words (i.e., neologisms such as **tasper* instead of *table*), and phonological paraphasias (e.g., **plower* instead of *flower*), false starts (e.g., *There is a d-d-d- dog*), sounds, and syllable repetitions. The term *words* refers to well-formed words produced with the exclusion of neologisms and phonological paraphasias. Each transcription was segmented in utterances. Utterance segmentation was carried out taking into account jointly the presence of clear pauses between utterances (*acoustic criterion*), the presence of

a complete semantic unit including a main predicate and its arguments (*semantic criterion*), the presence of a grammatically complete sentence with its subordinate clauses (*grammatical criterion*), and the presence of interrupted words or false starts (*phonological criterion*).

Narrative analysis was performed using a multilevel approach to micro- and macrolinguistic features of narrative production (Marini et al., 2011; Marini & Carlomagno, 2004). This procedure has been used in studies on cohorts of children with various clinical conditions (Marini et al., 2007, 2008, 2010), including school-aged children with ASD (Marini et al., 2020).

The microlinguistic analysis focused on three measures:

a) Mean length of utterance (MLU) calculated by dividing the total number of words by the number of utterances. Under the assumption that longer utterances require more words and, in principle, more complex grammatical characteristics, MLU provides indirect information about the grammatical skills of the participants.

b) Percentage of omissions of morphosyntactic information calculated by dividing the omissions of content words by the number of utterances and multiplying the result by 100. This percentage provides information about the participants' ability to adequately use the morphosyntactic information required by words while generating sentences.

c) Percentage of complete sentences calculated by dividing the grammatically complete sentences by the number of utterances and multiplying the result by 100. An utterance was considered a complete sentence if it did not contain any omissions or substitution of morphemes or words. Therefore, this percentage allows to directly assess the participants' ability to generate well-formed sentences.

The macrolinguistic analysis focused on three measures:

a) Percentage of cohesive errors calculated by dividing the number of cohesive errors by the number of utterances and multiplying the result by 100. A cohesive error included misuse of cohesive ties, such as connectives (e.g., *The man falls from the tree / but he hurts himself*), number and gender agreement between pronouns and nouns (e.g., *I saw John / and I told her about you*), and abrupt interruptions in an utterance, i.e., *aposiopesis* (e.g., *The man is... / The man falls from the tree*). This percentage provides information about the participants' ability to adequately link consecutive utterances by means of linguistic connectors.

b) Percentage of coherence errors calculated by dividing the number of global coherence errors by the number of utterances and multiplying the result by 100. Errors of global coherence included the production of utterances that were repeated (e.g., *There is a man / and... / There is a man*), fillers (e.g., *There is a... / I don't remember its name*), tangential (i.e., utterances with derailment in the flow of discourse, e.g., *The man falls from the tree / I really like trees*) or conceptually incongruent with the story. The percentage of coherence errors provides information about the speakers' ability to produce utterances that are adequately related to the main gist of the story and therefore of their discourse organization skills.

c) Percentage of lexical informativeness calculated as lexical information units (LIUs) in the narratives, such as content and function words that were not only phonologically well-formed but also grammatically and pragmatically appropriate (Marini & Urgesi, 2012). Words scored as errors of any kind and words embedded in fillers, repeated, incongruent or tangential utterances were excluded from the count of LIUs. The percentage of lexical informativeness was calculated by dividing the number of LIUs by the number of words and dividing the result by 100. The percentage of lexical informativeness provides direct evidence of the participants' ability to convey relevant pieces of information with their

words.

Transcriptions were performed by one coder while another coder transcribed 9 stories allowing for the calculation of the degree of reliability. The two coders reached almost perfect agreement based on the number of units ($ICC_{Average} = .996; p = .000$) and utterances ($ICC_{Average} = .946; p = .000$). Scoring was performed by the coder who has transcribed all the speech samples. A random sample of nine narratives was selected and given to an another coder for calculating interrater reliability. Overall, the two raters reached excellent agreement on % Complete Sentences ($ICC_{Average} = 1.00; p < .001$) Coherence errors ($ICC_{Average} = 1.00; p < .001$) and measures of MLU ($ICC_{Average} = 1.00; p < .001$) and almost perfect agreement on and cohesive errors ($ICC_{Average} = .925; p < .001$), measures assessing the omission of morphosyntactic information ($ICC_{Average} = .979; p < .001$) and lexical informativeness ($ICC_{Average} = .998; p < .001$).

Assessment of cognitive skills

Cognitive skills were assessed in one session of approximately one hour: the first assessment was conducted a few days before training began (T0); the second session was carried out immediately after training had finished (T1). The cognitive profile was determined using a selection of tasks of the Neuropsychological Evaluation Battery, standardized in Italian for pre-adolescents and adolescents (BVN 12-18; Gugliotta et al., 2009). See Table 3.2.

3.2 BRIEF DESCRIPTION OF THE NEUROPSYCHOLOGICAL TASKS ADMINISTERED PRE- AND POST- COGNITIVE-PRAGMATIC TREATMENT (GABBATORE ET AL., 2015)

Token test, 36 items (De Renzi & Faglioni, 1978)

The task assesses linguistic comprehension. The examiner reads a list of instructions of increasing difficulty regarding tokens differing in shape (squares and circles), size (large and small) and colour (green, white, yellow, red and blue). The first five sets of instructions are based on the verb 'touch', e.g., 'Touch a circle' or 'Touch the red circle'. The last set of instruction increases in difficulty and includes a wider variety of

actions, e.g., 'Before touching the yellow circle, pick up the red square'. Each instruction executed in a correct way is attributed a score of 1, while any instruction executed incorrectly is attributed a score of 0. The total score corresponds to the sum of the score obtained at each item (ranging from 0 to 36).

Naming task (Brizzolara et al., 1993)

The task assesses the ability to name items. The examiner shows, one by one, 88 black and white pictures, and asks the examinee to name them as fast as possible. The pictures present well-known objects belonging to the following semantic categories: animals, toys, tools, vegetables, cloths, fruits, pieces of furniture, means of transport, music instruments, domestic appliances, professions. The pictures' name may have high or low degree of iteration frequency in daily communicative interaction (e.g., chair vs. accordion), determining a certain variability in terms of complexity. Each pic correctly named is attributed a score of 1 while when a mistake occurs a score of 0 is attributed. The total score corresponds to the sum of the score obtained at each item (ranging from 0 to 88).

Digit Span and Corsi block-tapping test (Bisiacchi et al., 2005)

The tasks assess, respectively, verbal and spatial working memory. Specifically, they measure the ability to keep in mind a limited amount of information (numbers and locations/spatial relations between objects, respectively), in a readily available state, for a short period of time. In the *Digit span*, the participant is asked to repeat, after the examiner, sequences of numbers of increasing length. The total score is based on the longest series of numbers for which 2 or more sequences are correctly repeated. Score ranges from 0 to 9. In the *Corsi block-tapping test*, the examinee is presented with 9 wooden blocks arranged irregularly. The examiner taps the blocks in randomized sequences of increasing length (from 2 to 7), and the examinee is required to repeat the same sequence immediately after him/her. Each block-tapping series has three sequences of the same length. The total score is based on the length of the sequence of at least two taps (out of three) that the examinee repeats correctly. The score ranges from 0 to 7.

Immediate and Deferred Recall test for long-term verbal memory task (Spinnler & Tognoni, 1987)

The tasks assess the ability to extract and memorize information and recall it, immediately after its presentation and after a short time has elapsed. The examinee is required to repeat the content of a short text after listening to the examiner reading it out loud. The task is repeated once immediately after the examiner has read the text and again about 10 minutes later (in this time range the examinee is engaged in non-verbal tasks to rule out any possible interference with the present task). The content of the text is organized in main events (i.e., what has happened) and their secondary features (when and where), with different degree of relevance, which correspond to different scores. The total score is separate for immediate and deferred task and in both cases ranges from 0 to 8.

Selective attention (Bisiacchi et al., 2005)

The task assesses the ability to focus on a single or a few items in a given perceptual field, for a certain amount of time. The task material is made of a pattern of geometric shapes (i.e., squares with a line in different angles) displayed on a paper sheet. The examinee is shown the target square on the upper part of the sheet and, after a brief training, is required to mark all the squares on the paper sheet that look

exactly like the target one. Time limit is one minute. A score of 1 is given for each square correctly identified and the total score corresponds to the sum (range 0 to 21).

Tower of London (Shallice, 1982)

The task assesses planning ability. It requires the examinee to create a mental representation of the pattern of a set of given items and establish which actions are needed to switch from the baseline to the given goal configuration. The task is administered using a board with pegs and coloured wooden balls. The examinee is required, starting from an initial given configuration, to arrange 3 coloured balls on three upright sticks according to a series of given patterns pictured on a paper sheet. The instruction says to try to achieve the goal arrangement in as few moves as possible and by following simple given rules (e.g., do not move more than a ring at a time). A score of 1 is attributed each time the examinee sets the balls on the pegs according to the configuration given, within the maximum time of 1 minute and without breaking any of the rules. The total score corresponds to the sum of the scores attributed for each configuration (range 0-12).

Modified card sorting test (Nelson, 1976)

The task assesses shifting and inhibitory control and consists of 4 stimulus and 48 response cards displaying several symbols, different in colour (red, green, yellow, blue), number (1, 2, 3, 4), and type (triangle, star, cross and circle) of shape. The examinee is requested to sort the response cards so to place each of them below one of the stimulus cards. Each response card has only one feature in common with three of the stimulus cards, and none with the fourth one. The examinees are not given information about the sorting criterion to be used (i.e., shape, colour or number), but they are guided to discover the sorting rule at each move. A score of 1 is attributed for each criterion correctly identified and applied 6 times in a row. The total score represents the total number of categories correctly identified (range 0-8).

Assessment of Theory of Mind skills

Theory of Mind (ToM) abilities were assessed in one session of approximately one hour before (T0) and after the training (T1) by administering the following two tasks:

- The *Sally & Ann task* (Wimmer & Perner, 1983) was adopted for the assessment of first-order ToM. The task presents the examinees a false belief scenario with two paper dolls - Sally and Ann – acting as characters. The examinees are required to correctly interpret the characters' behaviour (looking for a ball in a box vs. in a basket), based on the beliefs attributed to the characters themselves (being aware or not of a location change). A correct interpretation receives a score of 1, an incorrect interpretation a score of 0.

- The *Reading the Mind in the Eyes test* (Baron-Cohen et al., 2001) was used for the assessment of ToM non mediated by language. The task is composed by 28 pictures portraying the eye region of several faces. After a practice item, the pictures are shown one at a time and the examinees are required to choose, among four possible alternatives, the word that best describes what the person in the picture is thinking or feeling. A score of 1 is attributed for each correct answer (0 otherwise). The total score is the sum of the score obtained at each item (0-28).

6.2.4. Data analysis

First, a Principal Component Analysis (PCA) with a direct oblimin rotation was conducted in order to explore the structure of the six narrative variables at T0 (pre-training) and T1 (post-training). Afterwards, participants' performance differences between T0 and T1 were assessed by comparing the narrative scores, cognitive and ToM measures with Wilcoxon tests for paired samples. Bonferroni's correction was used to adjust p-value threshold for multiple comparisons. Spearman's correlations of performance at T0 and T1 were performed to test for associations between narrative abilities. Bonferroni's correction was used to adjust p-value threshold for multiple correlations.

6.3. Results

6.3.1. Principal component analysis

The PCA was computed on the six narrative indexes at T0 and T1 with a direct oblimin rotation.

Concerning pre-training, the Kaiser–Meyer–Olkin measure verified the sampling adequacy for the analysis (KMO = .513). Bartlett's test of sphericity indicated that

correlations between variables were sufficiently large for PCA ($\chi^2 = 67.68$, $p < .001$). An initial analysis was conducted to obtain eigenvalues for each component in the data. Two components had eigenvalues over Kaiser's criterion of 1 and in combination described 80.86% of the variance. Given the convergence of the scree plot and Kaiser's criterion on two components, this is the number of components that were retained in the final analysis. The items that cluster on the same components suggest that component 1 represents the microlinguistic level and component 2 the macrolinguistic level.

Regarding post-training, the Kaiser–Meyer–Olkin measure verified the sampling adequacy for the analysis (KMO = .534). Bartlett's test of sphericity indicated that correlations between variables were adequate for PCA ($\chi^2 = 32.81$, $p < .005$). Eigenvalues for each component in the data were obtained. Two components had eigenvalues over Kaiser's criterion of 1 and in combination explained 68.31% of the variance. The convergence of the scree plot and Kaiser's criterion suggested to retain two components in the final analysis. The items that cluster on the same components suggest that component 1 represents the microlinguistic level and component 2 the macrolinguistic level. Data of both structure and pattern matrices in T0 and T1 are reported in Table IV (as suggested by Graham et al., 2003).

TABLE 3.3 PRINCIPAL COMPONENT ANALYSIS WAS CONDUCTED TO EXPLORE NARRATIVE DATA STRUCTURE IN T0 (PRE-TRAINING) AND T1 (POST-TRAINING).

| | T0 - Pre-training | | | | T1- Post-training | | | |
|--|-----------------------------|-------|------------------|-------|-----------------------------|-------|------------------|-------|
| | Pattern Matrix ^a | | Structure Matrix | | Pattern Matrix ^b | | Structure Matrix | |
| | Component | | Component | | Component | | Component | |
| | 1 | 2 | 1 | 2 | 1 | 2 | 1 | 2 |
| MLU | .858 | .194 | .868 | .235 | -.632 | .074 | -.625 | .013 |
| Omissions of morphosyntactic information | -.907 | -.071 | -.911 | -.114 | .893 | -.044 | .889 | .041 |
| Complete sentences | .875 | .054 | .878 | .096 | -.828 | .020 | -.826 | -.059 |
| Cohesive errors | -.666 | .432 | -.645 | .400 | .814 | .200 | .833 | .278 |
| Coherence errors | .323 | .890 | .365 | .905 | -.040 | .831 | .040 | .827 |
| LIUs | .108 | -.962 | .062 | -.957 | -.041 | -.895 | -.127 | -.898 |

Extraction method: Principal component analysis. Rotation method: Oblimin with Kaiser normalization. ^a Convergence for rotation performed in 5 iterations. ^b Convergence for rotation performed in 3 iterations.

6.3.2. Narrative assessment

The Wilcoxon tests for paired samples showed a significant improvement on two microlinguistic measures between pre- and post-training. The narratives contained longer MLU, and a higher percentage of complete sentences. Improvement on macrolinguistic measures was noted on the reduced occurrence of cohesive errors. It is worth noticing that omissions of morphosyntactic information, despite not being significantly improved, show a large effect size. However, a very similar percentage of global coherence errors and LIUs between T0 and T1 was reported (Table 3.4).

TABLE 3.4 PERFORMANCE PRE- AND POST- COGNITIVE PRAGMATIC TRAINING AT THE NARRATIVE ASSESSMENT.

| Linguistic level | T0 – Pre training | | T1 – Post training | | z | p* | r |
|--|--------------------------|---------------------|--------------------------|---------------------|-------|-------------|------|
| | Score range Min - Max | Raw score M (SD) | Score range Min - Max | Raw score M (SD) | | | |
| Micro | | | | | | | |
| MLU | 4.65-10.49 | 6.20 (1.39) | 5.33-12.93 | 8.12 (2.17) | 2.97 | .005 | .70 |
| Omissions of morphosyntactic information | 14.82-56.63 | 35.43 (13.50) | 10.10-39.33 | 24.39 (7.84) | -2.53 | .011 | .63 |
| Complete sentences | 25.54-68.21 | 48.24 (12.51) | 41.79-74.33 | 58.85 (8.24) | 2.84 | .004 | -.71 |
| Macro | | | | | | | |
| Cohesive errors | 5.00-60.89 | 25.22 (13.55) | .00-37.48 | 16.47 (10.43) | -2.79 | .005 | .70 |
| Coherence errors | .00-41.96 | 13.57 (10.21) | .00-25.22 | 10.83 (8.24) | -.91 | .363 | .23 |
| LIUs | 67.55-91.48 | 79.37 (7.80) | 69.14-87.64 | 77.96 (5.42) | -.67 | .50 | .17 |

Bonferroni's adjustment: $p > .008$; Significant p values are indicated in bold. z: standardized test statistics; r: effect size. MLU: Mean length of utterance; LIUs: Lexical Information Units.

Spearman's correlations on narrative variables in T0 (pre-training) showed a series of significant associations. MLU correlated with the percentage of omissions of morphosyntactic information, with the percentage of complete sentences, and with the percentage of cohesive errors. The percentage of omissions of morphosyntactic information correlated with the percentage of complete sentences. Finally, the percentage of coherence errors were statistically associated with LIUs. All other correlations were not statistically significant (see Table 3.5).

Spearman's correlations between narrative variables in T1 (post-training) revealed only one correlation between the percentage of omissions of morphosyntactic information and the percentage of cohesive errors. All others associations were not statistically significant although a moderate, but not significant, correlation is observable between the percentage of omissions of morphosyntactic information and the percentage of complete sentences. See Table 3.6.

TABLE 3.5 SPEARMAN'S CORRELATION ANALYSIS BETWEEN NARRATIVE ABILITIES ON PRE-TRAINING PERFORMANCE.

| | MLU | Omissions of morphosyntactic information | Complete sentences | Cohesive errors | Coherence errors | LIUs |
|--|--------------------------|--|-------------------------|-------------------------|--------------------------|------|
| MLU | - | | | | | |
| Omissions of morphosyntactic information | $r = -.70$ $p < .002$ | - | | | | |
| Complete sentences | $r = .69$ $p < .003$ | $r = -.90$ $p < .001$ | - | | | |
| Cohesive errors | $r = -.73$ $p < .001$ | $r = .39$ $p = .14$ | $r = -.50$ $p = .05$ | - | | |
| Coherence errors | $r = -.04$ $p = .89$ | $r = -.10$ $p = .71$ | $r = .06$ $p = .83$ | $r = -.02$ $p = .95$ | - | |
| LIUs | $r = .17$ $p = .53$ | $r = .08$ $p = .78$ | $r = -.05$ $p = .85$ | $r = -.14$ $p = .61$ | $r = -.94$ $p < .001$ | - |

* p -value threshold was adjusted for multiple comparisons with Bonferroni correction ($p < .008$). Degrees of freedom for each correlation = 14. MLU: Mean Length of Utterances. LIUs: Lexical Information Units

TABLE 3.6 SPEARMAN'S CORRELATION ANALYSIS BETWEEN NARRATIVE ABILITIES ON POST-TRAINING PERFORMANCE.

| | MLU | Omissions of morphosyntactic information | Complete sentences | Cohesive errors | Coherence errors | LIUs |
|--|--------------------------|--|-------------------------|-------------------------|-------------------------|------|
| MLU | - | | | | | |
| Omissions of morphosyntactic information | $r = -.37$ $p = .015$ | - | | | | |
| Complete sentences | $r = .47$ $p = .07$ | $r = -.56$ $p = .02$ | - | | | |
| Cohesive errors | $r = -.42$ $p = .11$ | $r = .74$ $p < .001$ | $r = -.46$ $p = .08$ | - | | |
| Coherence errors | $r = -.12$ $p = .66$ | $r = -.15$ $p = .58$ | $r = -.16$ $p = .54$ | $r = -.05$ $p = .86$ | - | |
| LIUs | $r = -.12$ $p = .65$ | $r = -.12$ $p = .65$ | $r = -.14$ $p = .61$ | $r = -.35$ $p = .18$ | $r = -.50$ $p = .05$ | - |

Bonferroni's adjustment: $p > .008$; Significant p values are indicated in bold. Degrees of freedom for each correlation = 14. MLU: Mean length of utterance; LIUs: Lexical Information Units.

6.3.3. Cognitive skills assessment

We compared the performance scores on the cognitive tasks administered pre- and post-training to determine whether differences could be found. As expected, there was no significant improvement in cognitive skills between pre- and post- training (Table 3.7).

TABLE 3.7 PERFORMANCE (RAW SCORES) PRE- AND POST- TRAINING AT THE NEUROPSYCHOLOGICAL EVALUATION BATTERY (ITALIAN STANDARDIZATION FOR PRE-ADOLESCENTS AND ADOLESCENTS, BVN 12-18).

| Task | T0 – Pre training | | T1 – Post training | | z | p* | r |
|---|--------------------------|---------------------|--------------------------|---------------------|------|------|-----|
| | Score range Min - Max | Raw score M (SD) | Score range Min - Max | Raw score M (SD) | | | |
| BVN 12-18 | | | | | | | |
| Token Test | 16-36 | 30.25 (5.42) | 27-36 | 31.31 (3.46) | .88 | .34 | .22 |
| Naming | 55-78 | 68.06 (6.79) | 60-84 | 72.19 (7.52) | 2.57 | .01 | .64 |
| Digit Span | 3-8 | 4.56 (1.21) | 4-7 | 4.62 (.96) | .26 | .79 | .07 |
| Corsi block-tapping | 4-6 | 5.25 (.77) | 4-7 | 5.44 (1.03) | .76 | .49 | .19 |
| Long term memory - immediate & delayed recall | 0-8 | 5.64 (2.52) | 3-8 | 6.25 (2.02) | .11 | .91 | .03 |
| Selective cancellation task | 1-20 | 11.31 (5.87) | 2-20 | 14.25 (5.00) | 2.42 | .015 | .61 |
| Tower of London | 4-12 | 9.12 (2.19) | 6-12 | 10.19 (1.72) | 1.91 | .06 | .48 |
| Modified Card Sorting Test | 1-8 | 5.31 (2.33) | 1-8 | 6.12 (2.30) | 1.44 | .15 | .36 |

Bonferroni's adjustment: $p > .006$; z: standardized test statistics; r: effect size.

6.3.4. Theory of Mind assessment

Participants' ToM scores were compared pre- and post-training in order to examine possible differences between the two observations. As previously hypothesized, no differences were detected between the ToM performances in pre- and post- A-CPT (see Table 3.8).

TABLE 3.8 PERFORMANCE (RAW SCORES) PRE- AND POST- TRAINING AT THE THEORY OF MIND TASKS

| Task | T0 – Pre training | | T1 – Post training | | z | <i>p</i> * | r |
|---|-------------------|--------------|--------------------|--------------|------|------------|------------|
| | Score range | Raw score | Score range | Raw score | | | |
| | Min - Max | M (SD) | Min - Max | M (SD) | | | |
| <i>Theory of Mind (ToM) assessment</i> | | | | | | | |
| Sally & Ann | 0-1 | .87(.34) | 0-1 | .87 (.34) | .00 | 1 | 0 |
| Reading the Mind in the eyes | 11-22 | 17.81 (3.21) | 12-22 | 17.75 (2.52) | -.29 | .77 | .07 |

Bonferroni’s adjustment: $p > .025$; Significant p values are indicated in bold. z: standardized test statistics; r: effect size.

6.4. Discussion

With the present study we investigated whether narrative ability in a cohort of verbally fluent adolescents with ASD could be improved after the administration of the Cognitive Pragmatic Treatment adapted for adolescents (A-CPT; Gabbatore et al., 2021). In its original version for adults, CPT has proven effective in improving the pragmatic skills of persons with traumatic brain injury (Bosco, Parola, et al., 2018; Parola et al., 2019) and schizophrenia (Gabbatore, Bosco, et al., 2017). The A-CPT has recently proved to be successful in increasing communicative-pragmatic skills in adolescents with ASD (Gabbatore et al., 2021), as assessed with the equivalent forms of the Assessment Battery for Communication (Angeleri et al., 2012, 2015; Bosco et al., 2012), in which the target variables investigated pertained the comprehension and production of a variety of pragmatic phenomena, expressed in different expressive means (including use of gestures and paralinguistic cues in addition to language per se). Narrative, the capacity to describe accounts of related events, is actually part of communicative ability; whereas some aspects of narrative are more linked to structural linguistic skills (phonology, etc.), other features (i.e., cohesion and coherence) are more connected to pragmatic inferential processes, such as implicatures (Cummings, 2014). Therefore, the aim of this study was to investigate whether the benefits provided by the A-CPT (a pragmatic training) can be extended to

another aspect of communication, i.e., narrative ability, through a different task (unlike A-CPT and ABaCo, the narrative task used in the present research is not based on the presentation of video clips). To the best of our knowledge, however, no data are available regarding the potential effects of such a treatment program on narrative production skills in adolescents with ASD. To fill this gap, we introduced before (T0) and after (T1) training the assessment of narrative production using a multilevel approach for the analysis of micro- and macrolinguistic aspects of narrative discourse (Marini et al., 2011). The within-group analyses showed a significant post-training improvement on two microlinguistic and on one macrolinguistic measure. These results will be discussed in light of previous studies on language development and functioning in persons with ASD.

Analysis of the narratives at the microlinguistic level showed a substantial increase in MLU and complete sentences from T0 (pre-training) to T1 (post-training). This improvement suggests that the greater grammatical efficiency, measured by the syntactic accuracy, contributed to enhancing MLU production. It is worth noticing that, despite not being statistically significant, omissions of morphosyntactical information approached significance, as observable also by the large effect-size, suggesting that also this variable underwent a sort of improvement with the training. MLU, omissions of morphosyntactic information, and syntactic accuracy were related to each other in the evaluation pre-training (T0), indicating that before the programme, longer utterances allowed for more complete sentences with less omissions of morphosyntactic information and, conversely, shorter utterances were linked to less complete sentences characterized by more omissions of morphosyntactic information. However, in the assessment post-training (T1), these associations between microlinguistic variables were no longer observable. This could be an effect of the training, which specifically improved some microlinguistic variables (i.e., MLU and syntactic accuracy).

Overall, these findings suggest that A-CPT may indirectly improve the morphosyntactic and grammatical skills of adolescents with ASD. This observation is shared by previous studies (Gillam et al., 2015; Petersen et al., 2014) that found improved the use of grammar by children with ASD after attending narrative treatment sessions focused on linguistic complexity (e.g., use of subordinating and coordinating conjunctions, adverbs, etc.) and story structure. Finally, a major component of A-CPT involves tasks devoted to strengthening pragmatic abilities expressed by linguistic means, such as conversation and role play. Since linguistic production was encouraged during the entire course of treatment, it is not surprising that some features of training, although A-CPT is pragmatically oriented, may have led to improvement on microlinguistic measures.

Concerning the macrolinguistic level of discourse processing, we observed a decrease in the percentage of cohesive errors in the post-training narratives. There was a significant negative correlation between this macrolinguistic variable and MLU in the assessment pre-training, suggesting that before receiving the training participants produced more cohesive errors and this affected the production of shorter MLU. Nevertheless, after treatment, the situation was different: the percentage of cohesive errors was not associated with MLU but it was positively correlated with the percentage of omissions of morphosyntactic information. This suggests that cohesive processing after training was likely influenced by an overall improvement in grammar. Concerning the percentage of global coherence errors and the percentage of LIUs, we observed no substantial improvement after training. This lack of improvement in the coherence index was somewhat surprising, as a previous study reported an improvement in narrative coherence (increased topic maintenance and a reduction in off-topic comments) in adolescents with ASD after a parent-mediated intervention that promoted the production of personal narratives in persons with high-functioning ASD (McCabe et al., 2017). However, the modality of intervention

and the assessment method in McCabe and co-workers' study (2017) differed from ours. For instance, the narrative intervention they described did not directly involve the adolescents with ASD but rather only their parents and caregivers. By contrast, the participants in our study were actively engaged in the training activities of the A-CPT programme. In addition, assessment of narrative discourse was performed using settings, tasks, and coding procedures that cannot be strictly compared between the two studies.

Regarding the percentage of LIUs, the absence of post-training improvement might be explained by the fact that A-CPT addresses a multimodal concept of pragmatics that comprises, in addition to language, other expressive means such as body gestures or tone of the voice. For our study, we focused on narrative ability expressed through language.

The difference in outcomes for cohesion, coherence, and LIUs suggests that these macrolinguistic features are to some degree independent of one another. For example, cohesion appears to be more related to the microlinguistic level, as supported by the association between cohesive errors and omissions of morphosyntactic information and also by the PCA results (for similar findings regarding participants with fluent aphasia, see also Andretta & Marini, 2015), whereas coherence and LIUs are likely to be more strictly related to the domain of pragmatics. In the assessment pre-training, the higher percentage of LIUs was related to a lower percentage of global coherence errors (as showed by the strong negative correlation). Admittedly, pragmatic ability covers a broad set of skills (Cummings, 2005). While some are directly targeted by the A-CPT program (e.g., the use of gestures, paralinguistic cues, social appropriateness), pragmatic features more closely related to discourse production are less emphasized during treatment. This might explain why these macrolinguistic features did not improve with treatment. In contrast, since language use was one of the expressive means targeted by the tasks in the A-CPT program, this might have enhanced not only microlinguistic discourse features but also narrative cohesion.

As regards cognitive assessment, in light of the complex and not fully clear interplay of cognitive functions and pragmatics in typical and atypical development (see Hyter, 2017; Matthews et al., 2018), a cognitive battery was administered before and after training to verify that its effect was specific for the target variable of the study (narrative ability) and not for the other variables investigated. As expected, we found no significant difference between T0 and T1 in the assessed cognitive measures with the exception of the naming task of the BVN 12-18 - which taps into lexical selection skills - that showed a significant trend toward improvement at T1. This result seems to support the specificity of the training for the target variable of the program, i.e., pragmatic ability.

Moving to the assessment of the ToM skills, we did not find any difference between T0 and T1 suggesting that, despite their still undefined connection, ToM and pragmatics address to independent abilities (Bosco, Tirassa, et al., 2018). Therefore, our results well exemplified the fact that pragmatic, and in this case also narrative, performance cannot be completely explained by the ToM ability otherwise an increase in ToM measures would have been observed in T1.

Overall, such results indicate a specific improvement in the target skills addressed by the training program, namely, communicative-pragmatic ability, rather than a general effect due to mere participation in social activities. Our data support the notion that pragmatic ability, which is also influenced by other cognitive functions, addresses something specific beyond the sum of other cognitive skills (Bambini, Arcara, et al., 2016; Bosco et al., 2019; Bosco & Gabbatore, 2017a, 2017b; Bosco, Tirassa, et al., 2018; Domaneschi & Bambini, 2020).

To our best knowledge, this is the first study to investigate the effect of A-CPT on the narrative abilities of adolescents with ASD, with a focus on micro- and macrolinguistic features of discourse production. Given the lack of studies regarding the effectiveness of

training to improve pragmatic and narrative skills in adolescents with ASD, our results fill this gap in the research. Also, the high average attendance rate (94.27%) suggests that the A-CPT sessions were perceived as enjoyable by our participants.

The present study has some limitations. First, although cognitive assessment served as a control measure, we did not compare our sample's performance to a control group of adolescents with ASD who did not undergo A-CPT. Future studies should include a control group to exclude the possibility of generic improvement. Also, normative data regarding narrative analysis for this age group are not available. This leaves open the question whether the baseline performance of our study sample was already so high that it precluded detecting pre- and post-training differences in their narrative performance. In this circumstance, normative data would be highly useful. While the structure of the narrative task minimizes any learning effect, for further studies it would be useful to develop equivalent forms of narrative stimuli so as to rule out any bias when adopting test-retest procedures. Even though in the present study we did not detect any learning effect as the cognitive performance scores remained overall stable in the two assessment phases, this is a factor that would deserve attention in future studies. The number of assessment tools for which equivalent/parallel forms are available is very limited; nevertheless, being able to control for practice and learning effects at different stages would be beneficial also for the cognitive tasks. In additions, the order of administrating the stimuli (i.e., of the stories) was not randomized. Since the pictures were shown in the same order to all participants, we were unable to control for this effect. Finally, future studies should focus also on visual narratives in order to investigate whether some pragmatic abilities are dependent or not from the expressive modality used (Adornetti et al., 2020).

In conclusion, this is the first study to focus on the improvement of narrative skills in adolescents with ASD after participation in a treatment program designed to improve their

pragmatic skills. This contributes to filling an important gap in the literature with potential impact on the occupational success, independent living, and community inclusion of persons with ASD, considering the importance of narrative skills for teenagers' educational achievement and psychosocial outcome. Our findings suggest that narrative difficulties may persist in ASD during adolescence and that a pragmatically oriented treatment program such as A-CPT may be useful in improving grammatical and cohesive efficiency on narrative discourse production tasks. The lack of statistically significant improvement in the ability to maintain overall coherence and to convey relevant pieces of information through words in the narratives suggests the need for treatments that improve the ability of children and adolescents with ASD to adequately plan, monitor, and produce samples of narrative discourse that are perceived as informative and communicatively appropriate by their interlocutors. This will be an area of focus in future research.

7. General discussion

The present doctoral thesis examines communicative ability, including pragmatics and narrative, in children and adolescents with pragmatic impairment caused by a physiological deficit or a neurodevelopmental disorder. Since the current literature is inconclusive on this matter, the present thesis takes hearing loss as example of physiological deficit, and age at first implantation as a predictor of pragmatic performance. Furthermore, given the scarcity of studies involving adolescents, this thesis focuses on autism spectrum disorder and on the assessment of the effects of training.

As regards HL, the final aim was to assess communicative-pragmatic abilities, considering first pragmatic language (*Study one*) and then other expressive means in addition to language, i.e., extralinguistic and paralinguistic elements (*Study two*), in school-aged children fitted with a cochlear implant for severe-to-profound (> 70 dB) hearing loss and to evaluate the role of the age at first cochlear implantation. Regarding ASD, the aim was to assess communicative-pragmatic ability, specifically narrative skills, in adolescents with ASD before and after a pragmatic intervention, i.e., the adolescents version of the Cognitive-Pragmatic Treatment (Gabbatore et al., 2021), and to collect data on improvement of these abilities also at a later age.

Concerning hearing loss, studies have reported mixed findings on the consequences of early cochlear implantation for the development of communicative-pragmatic ability (Guerzoni et al., 2016; Most et al., 2010; Nicastri et al., 2014; Rinaldi et al., 2013; Socher et al., 2019). The differences in outcome may be related to any of several variables: the type of assessment tools, the focus on pragmatic skills, and the age of participants. In addition, improvements and benefits associated with cochlear implants have been observed principally in the linguistic domain and most previous studies (Caselli et al., 2012; Church et al., 2017; Paatsch & Toe, 2014; Tye-Murray, 2003) have not considered other aspects that

are relevant in the pragmatic domain: extralinguistic and paralinguistic expressive means, appropriateness to the context, and compliance with social norms, for example, politeness in social contexts. Finally, the effect of age at early implantation on pragmatic performance of children who received a device has been rarely addressed in different age groups (Dammeyer, 2012; Toe et al., 2007). These open issues were the basis on which *Study one* and *Study two* were designed. The study results suggest that receiving a cochlear implant is fundamental for the development of communicative-pragmatic abilities comparable to those of individuals with normal hearing skills. As *Study one* shows, however, some deficits are still observable in the ability to understand and produce dialogue and to implement perspective-taking abilities in communicative ones in children with a cochlear implant. The results of *Study two* are shared by other findings on extralinguistic and paralinguistic abilities and their use in various contexts. Children with a cochlear implant display deficits in the ability to comprehend and produce additional cues that complement interaction, such as prosody, and in the capacity to produce communicative acts with respect to discourse (i.e., Grice's maxims) and social norms. Furthermore, these pragmatic deficits appear to decrease with age, except for metaphor comprehension, which is an ability typical of later development¹⁴. This exception could be due to the significantly poorer performance on tasks where perspective taking (traditionally linked to ToM) is recruited (i.e., Colours task): a recent longitudinal study showed that the ability to interpret metaphors and ToM development are interconnected (Del Sette et al., 2020). Finally, as reported in the literature, both studies show that improvement in communicative-pragmatic ability is enhanced by early cochlear implantation.

¹⁴ Unlike other communicative abilities that start to develop very early, such as adhering to a discourse topic or respecting turn-taking (Casillas & Frank, 2017), the use of metaphors requires finely developed complex linguistic and cognitive skills as prerequisite. Given the complexity of metaphors compared to literal language, they tend to be fully understood only in late childhood (Winner et al., 1976). Nevertheless, some recent studies suggest that at age 3-4 years, children start to understand some metaphors (Di Paola et al., 2020).

Previous studies on ASD have noted narrative impairment in individuals with the disorder (Baixauli et al., 2016; Carlsson et al., 2020). Narrative competence is an essential ability that helps individuals to create relationships and achieve satisfactory academic and professional goals. Efforts must be directed to improve treatment design and administration to improve the ability to communicate and interact. However, interventions are usually addressed to young children (Pico et al., 2021; Tam et al., 2021), whilst adolescents have been largely neglected so far (Parsons et al., 2017), except for two studies (Gabbatore et al., 2021; McCabe et al., 2017). This gap offers a starting point for *Study three* in which we showed that there is still room for improvement in narrative skills even at a later age. Adolescents with ASD experienced improvement of several components of their narrative abilities at the micro- and the macrolinguistic level after receiving cognitive-oriented pragmatic treatment (A-CPT; Gabbatore et al., 2021). No substantial improvement was noted in the mean length of utterance, the ability to produce a coherent story, and the quantity of informativeness. These findings, again, stress the need to design evidence-based interventions specifically for working on pragmatic abilities, including narrative skills.

In these three studies, communicative ability was shaped by two different interventions: a surgical one (i.e., cochlear implantation) and a cognitive one (i.e., A-CPT). Interventions are key to developing pragmatic and narrative abilities that can be comparable to individuals with typical development. Furthermore, results from *Study one* and *Study two* show that early intervention to improve pragmatic abilities is crucial because of the consequences for the development of effective social skills (e.g., Leonard et al., 2011) and general wellbeing (Haukedal et al., 2018). In contrast, results from *Study three* show that treatment at a later stage of development, such as adolescence, may still be effective and promote beneficial changes in narrative competence.

Some important issues arise in the context of the present dissertation. There is the need for accurate assessment of specific pragmatic and narrative difficulties that individuals with a cochlear implant and with ASD experience. Despite the large amount of empirical literature, inconsistent data lead to solutions that may be sufficiently helpful but not conclusive.

Existing rehabilitative programs should be enlarged to embrace and address pragmatic skills in which children with a cochlear implant appear to have difficulties, such as conversation, perspective-taking, and figurative language. Also, treatment should include newer strategies that have proven effective in empirical research, such as the beneficial effects of early exposure to other forms of communication, such as sign language. Interacting with other individuals using sign language before receiving a cochlear implant may foster the development of communicative-pragmatic ability (Hall et al., 2019; Rinaldi & Caselli, 2014).

Also, there is a need for treatments that increase the ability of individuals with ASD to organize and produce narratives that are perceived as informative and communicatively effective by their interlocutors. Future treatments must take into account the elements that promote social interaction in individuals with ASD, such as a predictable environment and technological devices. For instance, extensive empirical literature has shown that human–robot interaction is a promising methodology for developing the socio-communicative skills of individuals with ASD, since interacting with robots appears to encourage social engagement (Aryania et al., 2020; Dautenhahn & Werry, 2004; Kim et al., 2013; Zhang et al., 2019). This strategy can be employed in treatment protocols that include the use of a variety of technological devices, such as computers, and rehabilitative programs using tele-practice (Boisvert et al., 2010). Pragmatic interventions via tele-practice could significantly

increase the engagement of individuals with ASD and help them improve their pragmatic ability, including narrative skills, for better use in real-life situations.

Appendix A: ABaCo's Item examples¹⁵

(1) Linguistic Scale

Comprehension tasks

BSA: Assertion

Face-to face item #2:

Examiner: "That wall is painted in white."

Test question: Tell me if the sentence I told you is true or false.

Standard Communication Acts

Videotaped item #1:

Frank, Paula and Clare are in the kitchen, seated at the table, and they are having dinner. Frank says: "This pasta is very good, who has cooked it?" Paula answers: "I'm glad that my effort is appreciated".

Test question: Who cooked the pasta?

Nonstandard Communication Acts: Irony

Videotaped item #9:

In a shop, Lara tries on a dress that is clearly too tight and asks Simon: "Does this dress fit me?" Simon answers: "I see your diet works!"

Test question: In your opinion, what did the boy want to say to the girl?

If the participant repeats the actor's reply: What does it mean?

In-depth question: Was he serious?

In-depth question: Why did the boy answer in this way?

Production tasks

Standard Communication Acts

Videotaped item #13:

Husband and wife are sitting on the sofa. Wife: "What would you like to do this afternoon?"

Test question: What could the husband reply to the wife?

If the answer is not clear: What does it mean?

¹⁵ Part of these examples are taken from Angeleri et al.,2012.

Nonstandard Communication Acts: Deceit

Videotaped item #17:

Richard is in the bathroom. He inadvertently pours the perfume of his sister. He hastily dries and goes away. Sometime later his sister, with the empty bottle in her hands: “Who has poured my perfume?”

Test question: Richard doesn't want to be discovered. What could he answer?

If the answer is not clear: What does it mean?

(2) Extralinguistic Scale

Comprehension tasks

BSA: Request

Videotaped item #41:

The actress is standing on a chair next to a library and she is rearranging the books. A book falls from her hands and, looking at the camera, she points it as if asking the viewer to pick it up.

Test question: In your opinion, what did she want to say?

Nonstandard Communication Acts: Deceit

Videotaped item #53:

Henry is sitting at the desk with books and exercise books, but instead of studying he is watching TV. Suddenly he hears his mother coming and he switches off TV with the remote control. His mother comes in the room and looks at him with an interrogative expression as if asking “What are you doing?” Henry spreads his hands innocently, as if saying: “Nothing”.

Test question: In your opinion, what did the boy want to say to his mother?

If the participant repeats the actor's reply: What does it mean?

In-depth question: Was he serious?

In-depth question: Why did the boy answer in that way to his mother?

Production tasks

Standard Communication Acts

Videotaped item #59:

On a desolate country road, Deacon's car has broken down. Deacon seems to be there for a long time (he continuously watches at the clock, puts his hand over his eyes as if looking if someone is coming, he is nervous). Finally, he sees a car arriving.

Test question: The boy needs help. What gesture can be used?

If the answer is not clear: What does it mean?

(3) Paralinguistic Scale

Comprehension tasks

Basic Emotion

Videotaped item #77:

The actor is scared.

Test question: In your opinion, what emotion is he communicating? How does he feel?

- sad
- scared (target)
- happy
- calm

Paralinguistic contradiction

Videotaped item #80:

It's Robert's birthday. Monica gives him a gift. Monica: "Happy Birthday!" Robert opens the package and finds a tie with terrible colours. Showing bored face and voice, he says: "Thanks. Really, I really needed it... beautiful!"

Test question: In your opinion, what did the boy want to say to the girl?

If the participant repeats the actor's reply: What does it mean?

In-depth question: In your opinion, did the boy like the tie? Why?

Production tasks

Basic Speech Acts: Question, Command

Face-to-face item #1:

Examiner: "Give me the pen."

Test question #1: Try to ask me.

Test question #2: Try to order me.

Basic Emotion: Sadness, Joy

Face-to-face item #7:

Examiner: "Ask me where the doctor is."

Test question #1: Acting sad.

Test question #2: Acting happy.

(4) Context Scale

Comprehension tasks

Discourse norms—Grice's maxims (quantity)

Videotaped item #87:

Sister: "Where did you put my diary?" Brother, in front of a red chest of drawers: "In the red drawer."

Test question: Do you think the answer is correct?

If the patient says no: Why?

Social norms

Videotaped item #84:

The head office: "Miss, could you please type this letter?" The secretary replies with an angry tone: "Now I cannot! I have a lot of work!"

Test question: Do you think the secretary has been polite?

If the patient says no: Why?

Production tasks

Discourse norms

Face-to-face tem in vivo #2:

Examiner: "Imagine you are hungry and you are asking if dinner is ready

Test question #1: to a waiter

Test question #2: to one of you parents"

Test question: How would you ask?

(5) Conversational Scale

Topic #1 - Free-time

Face-to-face item #4:

Free conversation between participant and examiner.

Some suggestions for the examiner:

- What do you like to do in your free time?
- I really enjoy sport/reading/looking after my garden...

Depending on how the conversation proceeds:

- What sort of books do you like reading?
- Which sports do you like practicing?
- Which sports do you like watching on television?
- I am passionate about cars/football/stamp-collecting . . .
- Which team do you support?

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