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Pragmatic abilities of people with Williams syndrome

Bojana Arsic[a*], University of Belgrade, Studentski trg 1, Beograd, Serbia, bojana.arsic57@gmail.com, https://orcid.org/0000-0001-6728-3779
Anja Gajic[b], University of Belgrade, Studentski trg 1, Beograd, Serbia, anjuskagajic@gmail.com, https://orcid.org/0000-0003-2559-3669

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Abstract

This study reviews existing research on the pragmatic abilities of individuals with Williams syndrome. While previous literature describes linguistic strengths and weaknesses within this population, limited synthesis exists concerning pragmatic competence and its developmental patterns. Addressing this gap, this review aims to analyze contemporary findings to clarify specific pragmatic characteristics and identify directions for future research. Peer reviewed full text articles published in English between recent years were selected using systematic search procedures, and methodological quality was independently evaluated. Six studies met the inclusion criteria, comprising eighty participants with Williams syndrome. Findings reveal distinct pragmatic profiles characterized by assertive conversational behaviour, perseveration on topics, excessive verbal output, and difficulty shifting conversational focus. Evidence also indicates strengths in interpreting neutral facial expressions and particular emotional cues. Developmental trends suggest potential spontaneous improvement in pragmatic abilities with increasing age. All included studies demonstrated acceptable methodological rigor. The review concludes that pragmatic challenges among individuals with Williams syndrome require targeted assessment and structured intervention to support social communication. Recommendations include expanding sample diversity, standardizing assessment tools, and examining intervention effectiveness to improve pragmatic functioning.

***Keywords*:** Intervention; pragmatics; social communication; systematic review; Williams syndrome.

* ADDRESS FOR CORRESPONDENCE: Bojana Arsic, University of Belgrade, Studentski trg 1, Beograd, Serbia.
E-mail address: bojana.arsic57@gmail.com

1. INTRODUCTION

Williams syndrome (WS) or otherwise Williams-Beuren syndrome was first described in the 1960s (Williams et al., 1961) and is an autosomal dominant condition caused by the deletion of the seventh chromosome, which occurs with a frequency of 1:7500 (Metcalfe et al., 2005; Morris et al., 2020). As a consequence of this syndrome, problems with the cardiovascular and endocrine systems, as well as distinctive craniofacial features, often occur (Pober, 2010). In addition, these individuals have an accompanying intellectual disability (ID), as a result of which there are delays in speech-language development, difficulties in shifting attention, visuospatial memory, working memory, and planning (Frangiskakis et al., 1996; Mervis & Robinson, 2015; Morris et al., 2020; Pober, 2010; Rhodes et al., 2010; Barnhardt et al., 2024).

People with WS are hypersociable (Barak & Feng, 2016; Kozel et al., 2021; Niego & Benítez-Burraco, 2022; Nir & Barak, 2021), that is, they have a rich vocabulary, developed abilities of expressive language communication, excellent development of phonological processing abilities and verbal short-term memory, as well as a high degree of indiscriminate friendliness (Mervis & Velleman, 2011). In addition, people with this syndrome show a reduced sense of fear manifested through a lack of inhibition during contact with unknown persons (Crespi & Hurd, 2014; Järvinen et al., 2013), which leads to an increased risk of social rejection and abuse (Royston et al., 2019).

Despite developed communication abilities, people with WS show difficulties in the domain of pragmatics (Sepúlveda & Resa, 2024; Mervis & John, 2010; Mervis & Velleman, 2011; Tager-Flusberg & Sullivan, 2000; White, 2014). According to the American Psychiatric Association (2013), pragmatic communication disorder involves persistent difficulties when using verbal and non-verbal communication in the domain of social exchange of information, greeting, harmonizing communication with the context, respecting the rules and order in communication, and understanding the hidden meanings of linguistic constructions, with the absence of restrictive and repetitive interests. Pragmatic communication skills refer to the ability to use language in a social context. They include understanding the interlocutor's intentions expressed through verbal and non-verbal communication signals, understanding irony and sarcasm, understanding and respecting the context of communication, but also social norms (Carter et al., 2005; Norbury, 2014; Taguchi, 2011). Respect for social norms includes skills such as appropriate switching of interlocutors, interruptions, adequate selection of topics for conversation, use of eye contact, and other non-verbal strategies, all through the correct use of structural aspects of a particular language and respect for cultural aspects from which a person comes (Carter et al., 2005; Norbury, 2014; Taguchi, 2011).

Despite the hypersociability mentioned above, people with WS show difficulties in the domain of communication, such as difficulties in speech production, syntax, and understanding of more complex grammatical constructions (D'Souza et al., 2016). In the area of pragmatic abilities, people with WS have difficulties in the area of social cognition, understanding social signals, and maintaining and establishing friendships (Asada & Itakura, 2012; Morris, 2010), but show superiority in noticing emotional changes on the faces of interlocutors (Asada & Itakura, 2012; Lacroix et al., 2016; Van Herwegen, 2015).

Although some authors point out that pragmatic communication deficits can be difficult to measure using standardized tests because said deficits are manifested in the context in which communication takes place (Adams, 2002; Norbury, 2014; Volden et al., 2009), Đorđević (2023) points out how the assessment of

pragmatic abilities can be carried out to diagnose disorders, but also to determine the peculiarity of the pragmatic profile of a certain group of respondents.

1.1. Purpose of study

Therefore, this paper aims to review the research that shows the pragmatic abilities of people with WS.

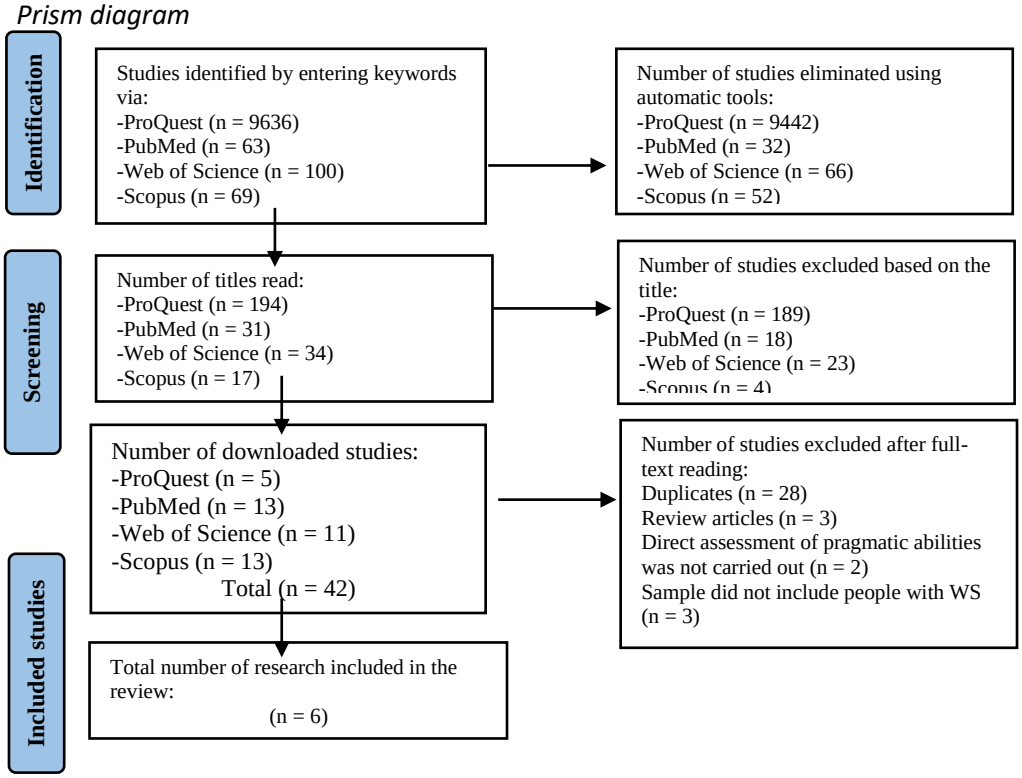
2. METHODS AND MATERIALS

The available literature was searched using the web search engines *ProQuest*, *PubMed*, *Web of Science*, and *Scopus*. The literature was searched using the following keywords in the English language: Williams Syndrome and pragmatics.

After searching based on keywords, using automatic tools, those articles that were published in full text, then articles that went through the peer review process, those that were published in English in journals in the form of an article, and in the period from 2017-2022. years were selected. The titles of the remaining papers were then read, based on which a further selection was made in the form of taking articles that, based on the titles, corresponded to the purpose of this paper. The received papers were then read in their entirety to determine whether they met the stated criteria, which included the fact that, in the research, a direct assessment of the pragmatic abilities of persons with WS was performed using some form of the assessment instruments. On that occasion, duplicates were excluded, followed by review papers, as well as papers in which no direct assessment of pragmatic abilities was performed, or those in which there were no persons with WS in the sample.

Figure 1

Prism diagram



Six papers were included in the final selection, and the selection process is shown in the Prism diagram shown in Figure 1. All papers included in the literature review were assessed for methodological quality using the *Quality Assessment Tool for Quantitative Studies* (Thomas et al., 2004). This instrument consists of eight domains, which relate to objectivity in sample selection, methodological design, confounding aspects of research, objectivity in measurement, methods of data collection, withdrawal of subjects from the research, integrity of the implemented intervention, and data analysis. Each of these domains has several items that direct the evaluator to what the research assessed with this instrument should satisfy, and each domain must be graded from one to three (1 = strong, 2 = moderate, and 3 = weak). Based on the individual scores for each of the domains, and based on the average, each study is assigned a quality score of strong (1.00–1.50), moderate (1.51–2.50), or weak (2.51–3.00). The author of this paper performed a quality assessment, followed by another independent assessor. Their grades were then compared to determine the concordance, as well as the reasons for the discrepancy, if any. Then a decision was made on whether there were too many discrepancies, and if there were none, the final quality score was calculated.

All the papers that met the inclusion criteria were further analyzed, concerning: a) respondents from the sample; b) instruments used in research; c) objectives of the conducted research; d) obtained results; and e) assessment of the quality of the mentioned studies.

3. RESULTS

Table 1 shows the papers included in the literature review, in alphabetical order, concerning the sample, the research objective, the instruments used in the research, the main results of the research, as well as the quality assessment of the study.

Table 1

Literature review

Reference	Sample		Research objectives	Pragmatic abilities	Instruments	Main results	QA
	Control group	Experimental group (WS)					
Alfieri et al., 2017	/	N = 32 M = 18, F = 14 AS = 12.3	Comparison of the influence of the mental age of respondents on pragmatic abilities, adaptive communication, and socio-communicative abilities	Receptive and expressive linguistic abilities Lexical and morphosyntactic understanding Lexical production Adaptive social skills	Leiter-R PPVT-r PVCL BNT	In the domain of adaptive socio-communicative abilities, chronological age has a greater influence than mental age on overall communication, lexical production, and lexical understanding Receptive communication is less developed than expected for the mental age In younger respondents, expressive communication and writing are significantly more developed than expected for their mental age In older respondents, communication follows mental age, and writing, receptive, and expressive lexical abilities are better than expected for mental age Writing improves with increasing chronological age	1.5

Diez-Itza et al., 2018	TP N = 8 M = 4, F = 4 AS = 5.5	N = 8 M = 4, F = 4 AS = 16.8	Assessment of the effectiveness of a short intervention on improving the pragmatic abilities of people with WS, based on the results of the assessment	Microstructural and macrostructural narrative retelling abilities	Narrative assessment based on watching a short silent cartoon	Microstructural aspects: Word production – on average 15.0 words with 26.13 clauses The complexity of the narration, on average 9.50 words in a sentence Macrostructural aspects: action recall with 65% accuracy on average, events with 37% accuracy on average, and characters with 87.50% accuracy on average	1.5
Diez-Itza et al., 2022	FXS N = 6 M = 6, F = 0 AS = 38.6	N = 6 M = 1, F = 5 AS = 23.11	A comparison of pragmatic abilities in four domains of individuals with FXS and WS	Comprehension and production of free speech	PREP	WS has a higher degree of topic perseverations, more excessive verbal production, word repetition, reformulations, word perseverations, and mixing of syntactic order compared to FXS	1.5
Perez et al., 2022	TP1 N = 40 M = 20, F = 20 AS = 3.3 TP2 N = 40 M = 20, F = 20 AS = 5.8	WS1 N = 7 M = 3, F = 4 AS = 5.8 WS2 N = 7 M = 5, F = 2 AS = 19.6	Presentation of pragmatic peculiarities of children and adolescents with WS in different domains of pragmatics	Phonological processing observed through syllable structure, substitution, omission, assimilation, and addition	RETAMHE	In all domains of phonological processing (syllable structure, substitution, omission, assimilation, and addition), the younger group of respondents with WS performed worse than the other groups of respondents, while there were no statistically significant differences between the other groups, which indicates a spontaneous improvement of the phonological index with increasing chronological age	1.3
Serrano-Juarez et al., 2020	DS N = 7 No data on gender AS = 12.57 TP N = 7 No data on gender AS = 11.14	WS is divided into 3 groups based on deletion type 1.1 Mb N = 1 M = 1, F = 0 AS = 14 1.5 Mb N = 7 No data on gender AS = 11 1.8 Mb N = 4 No data on gender AS = 12	Comparison of achievements between 5 groups (three groups with WS, DS, and TP) on the applied instruments	Social cognition through providing descriptions for desired behavior in specific situations, understanding facial expressions, and the theory of mind	WISC-IV WAIS-IV Eckman's faces TOM	People with WS with a 1.1 Mb deletion do best in the domain of social understanding The WS 1.1 Mb group is the best in identifying neutral faces, and the WS 1.8 Mb group in identifying fear. There was no difference between the groups in the identification of anger, joy, and sadness Theory of mind - 1.1 Mb group has the best achievements in theory of mind (total score on the applied instrument), but also first and second order theory of mind individually	1.5
Vandenheuvel et al., 2017	Deletion 22q11.2ds N = 8 M = 4, F = 4 AS = 9.6 (measurement 1)	N = 8 M = 5, F = 3 AS = 8.6 (measurement 1) AS = 10.4	Comparison of conversational skills between sample groups and comparison of progress in	Receptive and expressive communication in the aspect of structural communicatio	CELF-4-NL CELF-P2-NL WPPSI-III-NL Snijders-Oomen Test ALICC	Children with WS are more assertive than the other three groups Global conversational skills were statistically significantly better than the group of children with	1.5

	AS = 11.4 (measurement 2) ID N = 8 M = 7, F = 1 AS = 9.8 (measurement 1) AS = 11.4 (measurement 2) ASD N = 8 M = 6, F = 2 AS = 9.11 (measurement 1) AS = 11.7 (measurement 2)	(measurement 2)	the domain of conversational skills over time (longitudinal research)	n Reciprocity of communicatio n, taking into account the interlocutor's prior knowledge, maintaining the conversation topic, and discourse style	TOPICC	ID, but similar to children with ASD Children with WS have significantly more difficulties in changing the topic of conversation and discourse style than the group with ID WS is better than ASD in the domain of taking into account the interlocutor's prior knowledge during the conversation Children with WS, on the second measurement after some time, showed that they are getting worse in the domain of taking into account the prior knowledge of the interlocutor and that the deficit persists over time, while children with ID and ASD overcome it developmentally, or the deficit is stable

Note: QA – quality assessment; N – total number; M – male; F – female; AS – aritmetic mean; WS – Williams syndrome; TP – typicall population; ASD – autism spectrum disorder; ID – intellectual disability; FXS – Fragile X syndrome; DS – Down syndrome; WISC-IV – Wechsler Intelligence Scale; WAIS-IV – Wechsler Adult Intelligence Scale; Ekman's faces – Identification of facial emotions: TOM – Theory of Mind – Happe's Strange Stories; PREP protocol – Quick Protocol for Pragmatic Assessment; RETAMHE – Recording, Transcription and Analysis of Spontaneous Speech Samples; Leiter-R – Leiter International Performance Scale-Revised; PPVT-r – Peabody Picture Vocabulary Test-Revised; PVCL – Prove di Valutazione delle Competenze Linguistishe; BNT – Boston Naming Test; CELF-4-NL – Clinical Evaluation of Language Fundamentals – 4 – Dutch Edition; CELF-P2-NL – Clinical Evaluation of Language Fundamentals-2-Dutch Edition; WPPSI-III-NL – Matrix Reasoning and Picture Concepts Subtests of the Wechsler Preeschool and Primary Scale of Intelligence – Dutch Edition; SONR6-40 – Categories and Analogies Subtests from the Snijders-Oomen Nonverbal Intelligence Test; ALICC – The Analysis of Language Impaired Childrens Conversation; TOPICC – The Targeted Observation of Pragmatics in Childrens' Conversation Observation.

3.1. Sample used by the included studies

In the studies included in the literature review, 80 respondents with WS were included, of whom 37 were male and 32 female. For 11 respondents, the authors did not highlight data on gender. The average age of the respondents ranged from 5.8 to 23.1 years.

The largest number of studies from the literature review included in the sample children with WS (Alfieri et al., 2017; Perez et al., 2022; Serrano-Juarez et al., 2020), then persons with WS who were of adolescent age (Alfieri et al. al., 2017; Diez-Itza et al., 2018; Perez et al., 2022; Serrano-Juarez et al., 2021), while only one group of authors included adults with WS in their sample (Diez-Itza et al., 2022). Sauna-aho et al., (2019) found that in research conducted on a sample of people with any syndrome, people with different syndromes who are younger are most often included in the research. As potential reasons for fewer inclusion of adults with WS in research, it is highlighted that due to various health problems, they may have a shorter life expectancy (Collins, 2018; Nordstrøm et al., 2015), but also the loss of older people from the records (Fässberg et al., 2016), unless they are included in associations to help people with a certain

syndromic condition. Therefore, the only group that talked about the method of obtaining the sample was Serrano-Juarez et al. (2021). Namely, the authors pointed out that they acquired the sample by contacting the association to help people with WS. Considering that WS is not a syndrome that is common in the population and that there is not a large number of people with the mentioned syndrome in individual countries (Waz et al., 2021), this is expected.

When it comes to the group of subjects with WS, only two groups of authors from the literature review divided the sample of people with WS concerning the type of deletion they have and compared their achievements depending on the type of deletion the subjects have (Serrano-Juarez et al., 2021; Vandenheuvel et al., 2017). Individuals with one syndrome may have different types of gene deletion (Barak & Feng, 2016; Barak et al., 2019), which may affect the different expression of phenotypic features (Davies et al., 2020; Kehrer-Sawatzki et al., 2017; Reijnders et al., 2018; Shi et al., 2016).

In two studies from the literature review, the authors had as a control group a sample of people of a typical population (TP) who are of the same mental age (Diez-Itza et al., 2018; Perez et al., 2022), in one study (Serrano-Juarez et al., 2021) a control group of TP children also existed, but the authors included another one with Down syndrome (DS). One group of authors had people with Fragile X syndrome as a control group (Diez-Itza et al., 2022), while in the research of Vandenhavel et al. (2017), there were two control groups, i.e., one consisting of children with autism spectrum disorder (ASD) and the other of children with ID. In addition, only two groups of authors performed the equalization of the control and experimental groups of subjects by chronological and mental age (Serrano-Juarez et al., 2021; Vandenheuvel et al., 2017). Research shows that it is best to use as a control group a sample consisting of people of a TP of the same mental age to adequately observe the characteristics of people with a certain type of syndrome (Anderson et al., 2022; Bloomfield & Fisher, 2019).

Only half of the research authors included in the literature review testified to obtaining written consent for the inclusion of subjects in the research (Diez-Itza et al., 2018, 2022; Perez et al., 2022). Many authors emphasize the necessity of obtaining written consent from parents or guardians of persons with any type of ID (Ho et al., 2018; Strickler & Havercamp, 2023; Thompson, 2002), which includes persons with WS.

Only two groups of authors specified exclusion and inclusion criteria related to participant selection (Diez-Itza et al., 2018, 2022). In an earlier study by Diez-Itza et al. (2018), these were the presence of mild or moderate ID and the absence of comorbidity of ASD, while the same authors in their more recent study excluded those subjects with ID who had physical and sensory impairments present (Diez-Itza et al., 2022). It is of great importance to emphasize the selection criteria of respondents in the sample, because the presence or absence of certain accompanying conditions in respondents can have an impact on their performance (Meline, 2006).

To confirm the presence of WS in the subjects from the sample, most authors (Alfieri et al., 2017; Diez-Itza et al., 2018, 2022; Perez et al., 2022) performed a molecular genetic test called the FISH test (*Fluorescence In Situ Hybridization*), while only one group of authors performed the CMA test (*Chromosomal Microarray Analysis*). Both tests are applied in individuals who have different types of genetic mutations and allow the visualization of genetic peculiarities (Cui et al., 2016; Frickman et al., 2017; Huber et al., 2018).

Only Diez-Itza et al., (2018, 2022) reported in both of their studies that were included in the literature review how the respondents with WS from the sample spend their time, that is, what type of school they attend, in terms of their research conducted on a sample that included children with WS (Diez-Itza et al.,

2018), or the type of work the respondents do, when it comes to research by the same group of authors that included adult respondents (Diez-Itza et al., 2022). Davis (2007) points out that when describing the sample, it is necessary to provide relevant information about the respondents because they can give us an insight into the degree of functionality of the individual.

3.2. Data collection instruments used by the included studies

Đorđević (2023), in her article that focused on the categorization of instruments that can be used to assess pragmatic abilities, points out that the listed instruments are most often divided depending on whether discourse analyses, rating scales, analogical tasks, or test batteries are used. In almost all the studies included in the literature review, the assessment of the pragmatic abilities of people with WS included the assessment of discourse (Alfieri et al., 2017; Diez-Itza et al., 2018, 2022; Perez et al., 2022; Van Den Heuvel et al., 2017), while only in one study (Serrano-Juarez et al., 2021), respondents were given an analogous task to assess pragmatic abilities. Discourse assessment involves recognition of violations of social norms in conversation by the respondent (Đorđević et al., 2018), while analog assessment involves role-play in situations that are analogous to everyday interactions where the examiner observes the behavior of the examinee (Đorđević, 2023).

Of the studies above in which discourse was evaluated, in two studies (Diez-Itza et al., 2022; Perez et al., 2022) from the literature review, dialogue was evaluated. Evaluating the dialogue implies observation of the conversation in the everyday context (Đorđević, 2023). The evaluation of dialogues on various topics that are of interest to the respondents in the research from the literature review was carried out by evaluators, and the said conversations were then transcribed in both research and coded. Narratives were assessed in two literature review studies (Alfieri et al., 2017; Diez-Itza et al., 2018). Narrative assessment implies that the respondent talks about a certain topic or retells a story, based on the pictures he saw immediately or based on short stories read independently or by the examiner (Đorđević, 2023). When assessing the narrative of the respondents, Alfieri et al. (2017) showed the respondents photographs that they had to describe, while Diez-Itza et al. (2018) evaluated the narrative by showing a cartoon film to the respondents, which they recounted after watching it. The only research that evaluated discourse through exposure was the research of Vandenheuvel and colleagues (Van Den Heuvel et al., 2017). The authors asked the respondents questions following the *Test for the Assessment of Pragmatics* (The Analysis of Language Impaired Children's Conversation - TOPICC), after which they gave free answers based on their prior knowledge of certain topics.

In half of the studies included in the literature review, the authors assessed the intelligence quotient (IQ) of the respondents from the sample (Alfieri et al., 2017; Serrano-Juarez et al., 2021; Van Den Heuvel et al., 2017), however Van Den Heuvel et al., (2017) measured only non-verbal IQ, while the other two groups of authors (Alfieri et al., 2017; Serrano-Juarez et al., 2021) measured the total IQ of the subjects, to calculate the mental age of the subjects, and to equalize the control and experimental groups. It is of great importance to have data on IQ, as well as mental age, because the level of IQ can be a predictor of the level of development of other skills (Brawn et al., 2018), which is of great importance when it comes to people with WS, because their IQ can be in a wide range (McGrath et al., 2016; Twite et al., 2019), and may decrease over time (Fisher et al., 2016).

The most commonly used instrument for measuring IQ was the *Wechsler Intelligence Scale for Children and Adolescents - WISC-IV*, or the same instrument for adults (*Wechsler Adult Intelligence Scale - WAIS-IV*) (Serrano-Juarez et al., 2021; Van Den Heuvel et al., 2017), while only one study used other scales, such as the

Schneider-Oomen Nonverbal Intelligence Test (Vandenheuvel et al., 2017) and the *Leiter International Rating Scale-Revised* (LIRSC-r) (Alfieri et al., 2017). An IQ test is adequate for use if it is previously standardized, respects individual differences in the context of the previous level of education, and can also measure the practical application of intelligence in everyday situations (Kaplan & Saccuzzo, 2001; Kovacs & Conway, 2019; Sternberg, 2015).

A large number of research points out that the development of language skills is necessary for people to be successful in the domain of pragmatics (Baird & Norbury, 2016; Leech, 2016; Matthews et al., 2018; Urciuoli, 2016). This probably motivated half of the research authors included in the literature review (Alfieri et al., 2017; Perez et al., 2022; Van Den Heuvel et al., 2017) to assess the level of development of language skills in different domains, such as lexical skills (Perez et al., 2022), linguistic skills (Alfieri et al., 2017), and receptive and expressive structural language skills (Van Den Heuvel et al., 2017).

Of the additional assessment instruments, the only study from the literature review in which the authors used an instrument to assess the adaptive abilities of the subjects was the study by Alfieri et al. (2017). People with WS have less developed adaptive skills than people in the TP (Barak & Feng, 2016), and research shows that they decline with increasing chronological age (Morris & Mervis, 2021). Bearing in mind that individuals with WS can differ significantly in the degree of development of adaptive skills (Mervis et al., 2015), we assume that the mentioned group of authors considered this information to be important.

The only group of authors that evaluated the ability to recognize the emotions of others based on facial expression, as well as the theory of mind in the form of understanding strange stories in the respondents from the sample, was Serrano-Juarez et al. (2021). Theory of mind implies the ability to understand the mental state of another person (Imuta et al., 2016; Oakley et al., 2016; Schaafsma et al., 2015), and a large number of studies show that the degree of theory of mind development is a predictor of the development of pragmatic abilities (Andrés-Roqueta & Katsos, 2017; Bosco et al., 2017, 2018; Westra & Carruthers, 2017). In addition, research shows that children with various developmental and genetic conditions can be taught to recognize emotions (Cebula et al., 2017; Rice et al., 2015; Vasquez et al., 2015).

3.3. Research objectives of included studies

Two studies from the literature review aimed to compare the pragmatic abilities of WS and other syndromes (Diez-Itza et al., 2022; Serrano-Juarez et al., 2021), with the fact that in the study of Diez-Itza et al. (2022) was a group of subjects with Fragile X syndrome, while in the research of Serrano-Juarez et al. (2021), the sample consisted of a group of subjects with DS. Conducting research aimed at studying the characteristics of people with different syndromes in different areas is recommended because each syndrome has its characteristics (Carr et al., 2016; Vissers et al., 2016), and understanding the characteristics of individual syndromes can serve as a guideline in the treatment of people with different syndromes (Fletcher et al., 2018; Goering, 2015; Vissers et al., 2016).

Two studies were conducted to examine the impact of increasing mental or chronological age on the pragmatic abilities of people with WS (Alfieri et al., 2017; Van Den Heuvel et al., 2017), wherein one, the authors conducted a longitudinal study in which they studied the existence of spontaneous progress in the domains of pragmatics due to the increase in chronological age (Van Den Heuvel et al., 2017), and in the second, the authors examined the impact of the increase in mental age on pragmatic abilities (Alfieri et al., 2017). Research shows that people with WS have a poorer development of pragmatic abilities than expected for chronological age (Van Herwegen, 2015), but also, with increasing chronological age there can be

spontaneous development of different abilities (Morris & Mervis, 2021; Royston et al., 2019), while mental age was shown to be a predictor of the development of various abilities in people with WS (Alfieri et al., 2017).

Only one research from the literature review was focused on showing the pragmatic peculiarities of children and adolescents with WS in different domains of pragmatics (Perez et al., 2022), while also only one group of authors conducted a treatment aimed at improving the pragmatic abilities of children with WS (Diez-Itza et al., 2018). However, in our review, only the results related to the initial assessment, i.e., the pre-test conducted by the authors, are presented. A large body of research shows that the pragmatic abilities of people with WS can improve after the implementation of interventions in that domain (Royston et al., 2019; Sparks et al., 2019; Thurman & Fisher, 2015).

3.4. Research results of the included studies

In literature review studies that compared the pragmatic abilities of people with WS and other syndromes and developmental disorders (Diez-Itza et al., 2022; Serrano-Juarez et al., 2021; Van Den Heuvel et al., 2017) it was found that people with WS have better social understanding, theory of mind of first and second order compared to people with DS, but that they do not differ from people with DS in terms of understanding anger, joy and sadness, while people with WS are more successful in understanding neutral faces and fear (Serrano-Juarez et al., 2021). Research (Martínez-Castilla et al., 2015) conducted to identify the type of emotions that people with WS can recognize in others, and based on facial expression, showed that they more successfully recognize positive emotions in other people, while they have less developed ability to recognize negative ones.

When it comes to research that compared the pragmatic abilities of people with WS and Fragile X syndrome (Diez-Itza et al., 2022), people with WS show a higher degree of perseveration of topics during a conversation, more excessive verbal production, repetition, and perseveration of words, but also mix the syntactic order to a greater extent. Leon-Constantin et al. (2020) point out that the presence of perseverative content is the most common deficit in the conversation of people with WS.

By comparing the pragmatic abilities of people with ID, ASD, and WS (Van Den Heuvel et al., 2017), it was found that children with WS achieve better results in the domain of pragmatics than people with ID, initiate conversations more often and are more assertive than the other two groups of respondents (ID and ASD), show more difficulties in the area of changing the topic of conversation than children with ID, but take into account the interlocutor's prior knowledge during the conversation significantly better than the group with ASD. Previous research shows that people with ASD who have developed vocal communication skills have the greatest difficulties in the domains of conversational skills, as well as all the skills that conversation entails (Bambara, Cole, et al., 2018, Bambara, Thomas, et al., 2018; Sng et al., 2017; Ying Sng et al., 2018), so these results are not surprising. Therefore, they need teachers with great interest, patience, and skill to have effective communication and understanding (Uzunboylu et al., 2024; Uzunboylu et al., 2022).

Regarding the influence of the chronological and mental age of people with WS on pragmatic abilities, research from the literature review (Alfieri et al., 2017; Perez et al., 2022; Van Den Heuvel et al., 2017) concluded that with the increase in chronological age, children with WS have a spontaneous improvement in the area of phonological processing, and in the domains of syllable structure, substitution, omission, assimilation, and addition (Perez et al., 2022), as well as in the domain of written communication (Alfieri et al., 2017), while taking into account prior knowledge of interlocutor during conversation decreases with

increasing chronological age (Van Den Heuvel et al., 2017). In people with various developmental and genetic disorders, there is often a spontaneous improvement in a certain area of the deficit they exhibit, with an increase in chronological age (Ego et al., 2015; Zipper et al., 2017).

When it comes to mental age, receptive communication is less developed than expected, while expressive and written communication is more developed (Alfieri et al., 2017). In many syndromes accompanied by comorbid ID, abilities may be less developed than expected for their mental age due to great heterogeneity (Cleland et al., 2010; Hooper et al., 2008).

Research from a literature review that studied the narration of people with WS (Diez-Itza et al., 2018) showed that although people with WS produce a large number of words in a sentence, with difficulty accurately reproducing the events that happened, they show fewer problems when it comes to recalling the plot of an event, and they remember the characters best. Other research also comes to the result that people with WS have difficulties in recalling and reproducing events due to deficits in the domains of episodic memory (Mastrogiuseppe et al., 2019), as well as that when recalling events, they better remember people instead of events (Key & Dykens, 2016) due to increased focus on faces (Hsu & Ly, 2022; Jabbi et al., 2015).

The only study from the literature review that focused on comparing the achievement of subjects with WS who have different types of deletion (Serrano-Juarez et al., 2021) showed that, of the three groups of subjects with WS concerning the type of deletion, those subjects who have a 1.1 Mb deletion have the smallest deficits in the area of pragmatic abilities. It has been established that there are atypical and distinctive types of deletions in WS (Chailangkarn et al., 2016), as well as that there are differences in the degree of development of various abilities, depending on the type of deletion that a person with WS has (Barak & Feng, 2016; Kozel et al. al., 2021; Morris & Mervis, 2021).

3.5. Quality assessment of included articles

All studies included in the review were found to be of good quality (scores ranged from 1.3 to 1.5) when independently evaluated by two individuals. Both evaluators gave the same responses to 100% of the questions. Most of the articles included in the literature review (Alfieri et al., 2017; Diez-Itza et al., 2018, 2022; Serrano-Juarez et al., 2021; Van Den Heuvel et al., 2017) on quality assessment were assessed with 1.5 points, while only one study (Perez et al., 2022) obtained 1.3 points.

On the instrument used, in the domain of research design, the best assessment can be achieved if there is randomization when assigning respondents to the control and experimental groups, which is the only item that was fulfilled by the research of Perez et al., (2022), as a result of which the research of this group author achieved a better rating than other researches that were included in the literature review.

All studies included in the literature review (Alfieri et al., 2017; Diez-Itza et al., 2018, 2022; Perez et al., 2022; Serrano-Juarez et al., 2021; Van Den Heuvel et al., 2017) were assessed with a grade of two in the assessment of objectivity during the measurement. The reason for giving such a score is the lack of explanation to the respondents about the purpose of the research (or the lack of highlighting what was stated in the research itself). In addition, all studies from the literature review (Alfieri et al., 2017; Diez-Itza et al., 2018, 2022; Perez et al., 2022; Serrano-Juarez et al., 2021; Van Den Heuvel et al. ., 2017) on the domain related to withdrawal or removal of respondents from the research achieved a score of two, because the authors did not highlight relevant information in this regard.

4. CONCLUSIONS

Bearing in mind that people with different syndromes can have different pragmatic profiles and manifest different deficits in the domains of pragmatics, it is of great importance to conduct literature reviews related to individual syndromes, to show pragmatic abilities and characteristics of people with a certain syndrome, which is also the greatest importance of this work.

The results of this literature review indicate that people with WS exhibit different specificities in the domains of pragmatics, such as assertiveness, a high degree of perseveration of topics during conversation, and excessive verbal production characterized by the production of a large number of words in a sentence. The sentences they produce are characterized by repetitions and perseverations, mixing of syntactic order, and difficulties in changing the topic of conversation, but taking into account the interlocutor's prior knowledge and success in understanding neutral faces and fear. In addition, people with WS easily reproduce characters, while showing more pronounced deficits in terms of recalling the action of events, and with the increase in chronological age, there may be spontaneous improvements in the domain of pragmatics.

Although there can be a developmental improvement in the pragmatic abilities of people with WS, we emphasize the importance of implementing interventions in that domain, which we give as a recommendation for future researchers. Also, bearing in mind the diversity of deficits exhibited by people with WS with different types of gene deletions, as a recommendation for conducting future research, we highlight the determination of other specificities that do not exclusively include the domain of pragmatics.

No group of authors included in the literature review used a battery of tests to assess pragmatic abilities, and a large body of research shows that the application of such assessment instruments provides the most comprehensive results. As another recommendation, we point out the importance of applying test batteries in the assessment of the pragmatic abilities of persons with WS.

Bearing in mind the research quality ratings that were included in the literature review, as a recommendation for future researchers, we emphasize the necessity of randomization of sample groups, as well as highlighting the reasons for respondents' withdrawal from the research. Also, we highlight as a recommendation the importance of providing a clearer definition of the exclusion and inclusion criteria related to the selection of the sample, then information on obtaining written consent from the parents or guardians of the respondents, as well as the inclusion of respondents with WS in research that are of older calendar age.

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