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Title

Developmental course of conversational behaviour of children with 22q11.2 deletion syndrome and Williams syndrome

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Abstract (160 words)

This study investigated three conversational subskills in children with 22q11.2 deletion syndrome (22q11.2DS, *n* = 8, ages 7–13) and Williams syndrome (WS, *n* = 8, ages 6–12). We re-evaluated these subskills after 18 to 24 months and compared them to those of peers with idiopathic intellectual disability (IID) and IID and comorbid autism spectrum disorders (IID+ASD). Children with 22q11.2DS became less actively involved over time. Lower assertiveness than in children with IID was demonstrated. They seemed less impaired in terms of accounting for listener's knowledge than children with IID+ASD. Children with WS showed greater difficulties with discourse management compared to children with IID and 22q11.2DS. They had similar levels of conversational impairments to children with IID+ASD but these were caused by different shortcomings. Over time taking account of listener's knowledge became challenging for them. Findings suggest that children with 22q11.2DS and those with WS would benefit from conversational skills support and that regular re-evaluation is needed to anticipate conversational challenges.

Keywords

Williams syndrome, 22q11.2 deletion syndrome, conversation analysis, prospective longitudinal study, cross-syndrome comparison

Introduction

Conversation skills are considered an essential medium through which children establish cooperative relationships, which can influence social acceptance, and which may lead to social rejection or isolation from peers (Hemphill & Siperstein, 1990). Responsiveness and participation in daily conversations allow children to establish and maintain cohesive social interactions and thus characterise their social status (Black & Hazen, 1990). Children who have difficulties with interacting in a naturalistic context such as a conversation, are known to experience difficulties with peer relationships and have a higher risk of being bullied than typically developing children (Murphy, Faulkner, & Farley, 2014; Conti-Ramsden & Botting, 2004).

Communication subskills that may contribute to a successful conversation, include the following: (1) the ability to take into account listener's knowledge, (2) discourse topic management, and (3) responding contingently and extend a topic by providing relevant information on a conversational partner's turn (Black & Hazen, 1990; Nadig, Lee, Singh, Bosshart & Ozonoff, 2010; Schegloff, 2000). The refinement of each of these conversational skills encourages children's communicative effectiveness (Baines & Howe, 2010; Dorval & Eckerman, 1984)

Development of conversational subskills

The development of conversational abilities is a gradual process and allows children to become competent speakers in a complex and dynamic communicative environment (Short-Meyerson & Abbeduto, 1997). We discuss the development of the three abovementioned subskills in turn.

By 4 years of age, typically developing children are able to skilfully adapt to their conversational partners (Dewart & Summers, 1995). They make assumptions about the beliefs, knowledge and intentions of other people in order to appropriately select and express speech acts. Accurate judgment of the listener's informational needs relies on perspective-taking and role-taking abilities which advance considerably from 6 to 9 years of age (e.g. Clark & Svaib, 1997; Lloyd, Camaioni, & Ercolani, 1995; Lloyd, Mann, & Peers, 1998). Furthermore, taking account of listener's knowledge has found to be closely related to children's developing theory of mind (Cummings, 2013). Hence, a close relationship between this conversational proficiency subskill and the development of social cognition has been suggested (McTear & Conti-Ramsden, 1992; Whitehurst & Sonnenschein, 1985). Short-Meyerson and Abbeduto (1997) reported that even young children (4 to 5 year olds) applied strategies to assess and adapt to their discourse partner's knowledge. The development of socio-cognitive skills allows children to reason about the thoughts and knowledge of others, and to accommodate their interactions accordingly (Short-Meyerson & Abbeduto, 1997). Furthermore, children's inhibitory control skills facilitate the inhibition of their own perspective and enable them to take listener's perspective into account (Nilsen & Graham, 2009).

In addition, increasing participation in discourse supports children in using a variety of devices to introduce and maintain topics (Leinonen, Letts & Smith, 2000). Overall typically developing children seem to be motivated to initiate social interactions even before they acquire language. During preschool and primary-school years initiations become more sophisticated and children become able to produce informative messages that are appropriate given the conversational context (McTear & Conti-Ramsden, 1992).

Directives support the pacing of talk and turn-taking. Therefore, they play a crucial role in the emergence of topic coherence. Whilst children of 7 to 8 years old will still make unrelated and tangential contributions (about 37%), this proportion significantly decreases in children of 10 to 11 years old who return to topic more often (Dorval & Eckerman, 1984). Discourse topic management has also been linked to an increasingly mutual and active participation level, to the ability to account for an alternative perspective, and an increase in questioning of the conversation partner's justifications and thinking (Baines & Howe, 2010).

A third subskill enhancing conversational success is conversational responsiveness and the willingness to elaborate on the ongoing topic. Discourse is a joint activity and can only exist through accommodation on both sides (Perkins, 2007). Reciprocal verbal exchange develops alongside with the acquisition of structural and pragmatic language proficiency (Nadig et al., 2010). Beyond the infant stage, children will improve their ability to sustain in longer and longer sequences of turns (Dewart & Summers, 1995). The ability to maintain a topic also significantly increases in early typical childhood (e.g. Baines & Howe, 2010; Short-Meyerson & Abbeduto, 1997).

Conversational skills in children with intellectual disability

Children with intellectual disability (ID) encounter various challenges in the development of the aforementioned conversational subskills. Firstly, they experience difficulties in formulating ideas in which the referents are clear to the listener (Brownell & Whiteley, 1992). They also seem to have challenges with adapting their utterances to the characteristics of the listener (Kamhi & Masterson, 1989). Since taking listener's knowledge into account relies on both linguistic and socio-cognitive abilities, children

with ID have an increased risk of difficulties in this domain (Hatton, 1998; Rondal, 2001). Research has revealed limitations in perspective- and role-taking abilities and in speech act expression in heterogeneous groups of children with ID. (Abbeduto & Hesketh, 1997; Short-Meyerson & Abbeduto, 1997). Nevertheless, most children with ID produce utterances that are appropriate given the topic, even though the quality of their contributions may vary (Abbeduto & Hesketh, 1997).

Conversational skills in children with ID and comorbid autism spectrum disorders

Pragmatic language impairments including a range of conversational shortcomings are more pronounced in children who are diagnosed with ID and a comorbid autism spectrum disorder (ID+ASD). Lack of conversational initiation, flexibility and social engagement in interaction have been reported in these children (McGee, Feldman, & Morrier, 1997; Muskett, Perkins, Clegg, & Body, 2010). Furthermore, conversational cohesion is disrupted due to inappropriate, minimal, or vague contributions and unbalanced turn-taking (Fine, Bartolucci, Szatmari, & Ginsberg, 1994; Tager-Flusberg & Anderson, 1991). Impaired responses and profound comprehension difficulties cause conversational breakdowns (Asberg, 2010; Capps, Kehres, & Sigman, 1998). Some studies have indicated gains over time in topic management, pragmatic language and non literal abilities (Hale & Tager-Flushberg, 2005; Whyte & Nelson, 2015). Additional longitudinal cross-syndrome studies are necessary to draw more accurate conclusions. This is especially true when considering whether children with ID+ASD acquire conversational skills in a different way compared to groups with other neurodevelopmental disorders.

Cross-syndrome research on conversational skills

Increasing interest in the aetiological dimension of neurocognitive and behavioural variability observed in children with ID goes alongside continued advances in genetic and molecular techniques (Rondal, 2001). Language studies in children with genetic disorders (e.g. Fragile X, Down syndrome, Williams syndrome) have supported the idea of syndrome-specific pragmatic characteristics and socio-communicative challenges (e.g. Levy, Tenenbaum, & Ornoy, 2000; Price et al., 2008; Stojanovik, 2006).

Two groups whose communicative contributions are also characterised by several pragmatic deficits are those with 22q11.2 deletion syndrome (22q11.2DS; Antshel, Marrinan, Kates, Fremont, & Shprintzen, 2009) and those with Williams syndrome (WS; Brock, 2007). Antshel et al. (2007) described that 41% of children with 22q11.2DS met liberal criteria for autism on the Autism Diagnostic Interview-Revised. A comorbid diagnosis of ASD was indicated to be present in 23% of children with 22q11.2DS (Niklasson, Rasmussen, Oskarsdottir, & Gillberg, 2009). For WS, differences from and similarities to the social phenotype of children with ASD have also been reported (Asada & Itakura, 2012). Therefore, children with 22q11.2DS and WS are likely to be at risk of problems in the area of conversational interaction. The present research focuses on these two microdeletion syndromes and we therefore provide a concise review of their pragmatic and conversational challenges.

Conversation skills in children with 22q11.2 deletion syndrome

22q11.2 deletion syndrome (22q11.2DS) occurs in approximately 1:4000 births (McDonald-McGinn et al., 2015). The syndrome is associated with a broad spectrum of

cognitive, learning, motor and communicative disorders (McDonald-McGinn & Sullivan, 2011).

Until now, pragmatic language characteristics, and more specifically conversational competences, have received little attention in this group, but there are some important exceptions. Parents of children with 22q11.2DS have reported concerns regarding (1) inappropriate information transfer (2) difficulties with initiating conversations, and (3) neglect or inadequate use of contextual cues and turn-taking difficulties (Angkustsiri et al., 2014; Van Den Heuvel, Manders, Swillen, & Zink, 2017). These concerns have been confirmed in experimental studies. Impoverished information transfer and difficulties with initiating a story retelling were reported in 18 Swedish speaking children (5–8 years old; Persson et al., 2006). Van Den Heuvel and colleagues (2016) also found ambiguity to be a feature of language in 27 school-aged children with 22q11.2DS. They reported inadequate use of contextual cues, subsequent difficulties in selecting appropriate speech acts during a role-taking task and an elevated number of irrelevant or off-topic elaborations in a barrier-game. (Van Den Heuvel et al., 2016). Other information on conversational features in children with 22q11.2DS comes from anecdotal descriptions rather than systematic research. Solot et al. (2001) reported that disorganised discourse was frequently observed in school-aged children. In another study, poor responsiveness to simple questions and withdrawal behaviour during conversations was noted in children with 22q11.2DS aged 3 to 6 years of age (Golding-Kushner, Weller & Shprintzen, 1985).

Conversational skills in children with Williams syndrome

Williams syndrome (WS) is a fairly rare microdeletion syndrome (1:7.500 live births) (Strømme, Bjornstad, & Ramstad, 2002). In contrast to their mild-moderate cognitive impairments, children with WS have relatively spared structural language abilities in line with their nonverbal level of functioning (e.g. Karmiloff-Smith, Klima, Bellugi, & Baron-Cohen, 1995; Mervis & Velleman, 2011).

Despite the willingness of children with WS to engage with others, several pragmatic language challenges are likely to cause communicative breakdowns (e.g. Laws & Bishop, 2004; John, Dobson, Thomas, & Mervis, 2012). Firstly, due to poor judgment of the listener's informational demands, children with WS have been found to provide too little information in conversation (Stojanovik, Perkins, & Howard, 2001; Tarling, Perkins & Stojanovik, 2006). Secondly, the initiation of conversations of individuals with WS is often considered insistent and inappropriate (Laws & Bishop, 2004). They have been reported to chatter ceaselessly, ask socially inappropriate questions and to repeatedly use stereotypical phrases. A third characteristic of their conversational exchange is a high number of extended and inadequate responses (Skwerer et al., 2011; Skwerer, Ammerman, & Tager-Flusberg, 2013), and a decreased use of continuations. These issues result in a less successful flow of conversation highly dependent on the lead and contributions of their interlocutor (Lacroix, Bernicot, & Reilly, 2007; Stojanovik, 2006). Associated with this conversational imbalance, many children with WS have problems with establishing friendships and experience social difficulties leading to isolation (Klein-Tasman, Li-Barber, & Magargee, 2011; Riby et al., 2014). A large proportion of existing research has applied questionnaires to report on pragmatic strengths and challenges in children with WS. A more in-depth direct analysis of their conversational style and

pragmatic impairments might confirm and elucidate the previous (indirect) findings. A comparison to other groups of children with neurodevelopmental disorders could also provide an avenue for more individualised syndrome-specific pragmatic language intervention.

Current study design and aims

In summary, the existing literature lacks studies examining whether the discourse skills of children with 22q11.2DS and WS differ from those of children with idiopathic intellectual disability (IID) and of children with IID and comorbid ASD (IID+ASD). Further studies are needed using both (1) a quantitative analysis after coding of conversational turns, and (2) a categorical analysis at a global level evaluating the conversation as a whole to characterise conversational shortcomings in children with neurodevelopmental disorders. Finally, exploring developmental changes may reveal an atypical course of conversational behaviours in children with 22q11.2DS and WS. This might have important implications for assessment and may lead to syndrome-specific recommendations.

The present study used a cross-syndrome comparison and a prospective longitudinal follow-up design to detect possible subtle differences across groups of children with neurodevelopmental disorders. The aim of the research was to examine children's (1) ability to take listeners knowledge into account, (2) discourse management skills, and (3) responsiveness and elaboration to the ongoing verbal exchange. All of these conversational subskills were assessed twice during middle childhood by means of (a) participation and assertiveness indexes following Bishop, Chan, Adams, Hartley & Weir (2000), and (b) the Target Observation of Pragmatics in Children's Conversation

(TOPICC; Adams, Gaile, Freed, & Lockton, 2010), a four-point scale to evaluate conversational behaviours at a global level in several subcategories. Emphasis was on the following questions:

1. Do conversational subskills (i.e. taking account of listener's knowledge, discourse topic management, elaborations on a partner's turn) of children with 22q11.2DS and children with WS differ from those of age-matched children with IID or children with IID+ASD?

2. Is there a difference in the developmental changes (i.e. Time 2 minus Time 1, difference scores) of conversational competence, measured by means of changes in number of utterance and responses, assertiveness and TOPICC scores across groups?

3. Are there differences in conversational skills of children with 22q11.2DS and children with WS?

Methods

Participants

Conversational behaviours of 33 children were assessed at two time points. Four groups of children (22q11.2DS, WS, IID and IID+ASD) participated in the study.

Children with 22q11.2DS (n = 8) and WS (n = 8) were recruited from (Center for Human Genetics, University Hospitals Leuven)*).* Inclusion criteria were: (a) confirmed diagnosis of 22q11.2DS or WS by means of fluorescence in situ hybridisation or microarray technology, (b) presence of cognitive impairment indicated by at least one standardised intelligence assessment prior to the study resulting in FSIQ < 85 (i.e. -1 *SD* below the mean).

Children with IID (*n* = 13) were ruled out when any known genetic anomaly was reported in the parents' survey or was found in their medical record or was suggested after examination by a medical doctor. In all but the IID+ASD group (*n* = 12), ASD diagnosis was an exclusion criterion for the present investigation.

Autism spectrum disorder (ASD) was diagnosed according to the *Diagnostic and Statistical Manual of Mental Disorders* 4th edition criteria (DSM-IV; American Psychiatric Association, 1994) after a broad child psychiatric assessment using the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 1999) and/or Autism Diagnostic Interview – Revised (ADI–R; Rutter, Le Couteur, & Lord, 2003). Exclusion criteria for all groups were: prematurity (i.e. birth before 37 weeks) and severe sensorimotor deficits (bilateral hearing loss ≥ 40 dB HL or corrected visual acuity below 20/40). Only monolingual Dutch-speaking children were enrolled in the study.

Structural language and IQ measures

Language proficiency and intellectual functioning were examined in three to four sessions of approximately one hour each. Receptive and expressive structural language skills were evaluated by means of the Clinical Evaluation of Language Fundamentals 4–Dutch edition (CELF–4–NL; Kort, Compaan, Schittekatte, & Dekker, 2010) or the Clinical Evaluation of Language Fundamentals–Preschool–2 Dutch edition (CELF–P2–NL; de Jong, 2012) depending on the chronological age of the child.

The first author (EVDH) examined the language abilities of all children with WS and 22q11.2DS. Four trained research assistants (from the Master in Speech, Language and Hearing Sciences degree at KU Leuven) collected data from the control groups (IID and IID+ASD, *n* = 25). The Core Language Score (CLS; *M* = 100, *SD* = 15) of the

CELF–P2–NL (de Jong, 2012) or the CELF–4–NL (Kort et al., 2010) was calculated to assess the overall structural language level of the children.

At the first time point the CLS of children with 22q11.2DS tended to be higher than the CLS of children with IID (Table 1). At the second time point, it was found to be significantly higher in the 22q11.2DS group. In particular, sentence production (formulating sentences score, FSS) of children with 22q11.2DS was significantly higher compared to children with IID (Table 1).

INSERT TABLE 1

The overall language level of children with WS corresponded well to that of children with IID at both time points. At the second time point children with WS tended to have poorer sentence comprehension scores (SCS) compared to children with IID but no significant group difference was demonstrated. All children with WS had sentence comprehension scores of at least two standard deviations below the mean at the second time point (Table 2). The language proficiency of children with 22q11.2DS matched the language proficiency of children with WS well (U = 27.50, p = .668). Children with WS and 22q11.2DS were less well matched for CA (U = 24.00, p = .425) and for NVMA (U = 22.00, p = .314) at Time 1. It should be taken into account that children with WS were slightly younger and had a slightly lower NVMA when interpreting the cross-syndrome results.

INSERT TABLE 2

Cognitive assessments were supervised by the fifth author (AS). Five broad cognitive abilities were evaluated within the framework of the Cattell-Horn-Carroll model (Newton & McGrew, 2010). Averaged age equivalents of nonverbal fluid reasoning subtests (G*f*

index), defined as nonverbal mental age, were used as a matching criterion. In children between 5 and 8 years of age Matrix Reasoning and Picture Concepts subtests of the Wechsler Preschool and Primary Scale of Intelligence Dutch edition (WPPSI–III–NL; Hendriksen & Hurks, 2009) were used for measuring G*f*. In children aged 8 years and above, G*f* was evaluated by means of the Categories and Analogies subtests from the Snijders-Oomen Nonverbal Intelligence Test (SON R6–40; Tellegen & Laros, 2011).

Matched design

Children with microdeletion syndromes were matched for chronological age (CA) and nonverbal mental age (NVMA) to a group of children with IID and to a group of children with IID+ASD at Time 1. This resulted in a cohort of eight children with 22q11.2DS and eight children with WS matched to in total 25 children with IID or IID+ASD. In general, different individuals with IID and IID+ASD were used as controls for the 22q11.2DS group and WS group. However, there was some overlap, such that seven children were a control for both a child with 22q11.2DS and for a child with WS.

Kruskal-Wallis tests demonstrated that children with 22q11.2DS were well matched for CA to the IID and IID+ASD groups ($H(2) = .005$, $p = .997$) and also for NVMA ($H(2) = .590$, $p = .745$) at Time 1. Children with WS were also well matched for CA ($H(2) = .139$, $p = .933$) and for NVMA ($H(2) = .039$, $p = .981$) to both control groups at Time 1. Post-hoc Mann-Whitney *U* comparisons (Table 1 and 2) confirmed that groups were well matched at Time 1 ($p > .500$, Frick, 1995). At Time 2, the 22q11.2DS group was no longer well matched for NVMA to the IID group (Table 1). Children with WS were no longer well matched for NVMA to the IID+ASD group at Time 2 (Table 2).

Conversation analysis

Conversational data were collected at two time points with an interval of 18 to 24 months. The conversation task was always administered after the structural language assessment in the last session. This was intentionally chosen to allow children to get acquainted with the assessment environment (video-recording) and the examiner. All assessments took place in the child's school or at their home in a quiet room. All parents were individually informed about the aims of the research project, signed a consent form and participated voluntarily. The study protocol was approved by the Institutional Review Board of the KU Leuven and University Hospitals Leuven. .

Elicitation. The 'Analysis of Language Impaired Children's Conversation' (ALICC) procedure described by Adams and Bishop (1989) was applied. During the conversation task the examiner introduced three topics by means of pictures, put these aside, and encouraged the child to talk about similar experiences. Three coloured pictures were selected from 'Colour Cards ® Emotions / Sequences in 6-8 steps for Children' depicting every day, recognisable situations to initiate the conversation. Detailed information regarding the situations depicted in the pictures is provided in Appendix 1. At Time 1, the first author (EVDH) was the interlocutor of all children with 22q11.2DS and WS. As for the standardised language tests, the four research assistants collected data from the control groups (IID and IID+ASD, n = 25). At the second time point, the conversations of all participants were guided by the first author. The goal was to collect ten minutes of conversation in which the examiner elicited either information or an expression of involvement. One hundred conversational turns were sampled. Medians and ranges of total number of utterances are displayed in Tables 4 and 5.

Transcription. All samples were transcribed using Systematic Analysis of Language Transcripts conventions (SALT; Miller, Andriacchi, & Nockerts, 2011) for each individual after ignoring the first ten conversational turns, which were considered as 'warm-up'. A turn consisted of utterances that passed from one interlocutor (Examiner/E) to the other (Child/C).

Coding and measures. Firstly, the framework of Adams and Bishop (1989) was used and a subset of coding categories was selected based on the previous studies of Adams and Lloyd (2005), Bishop, Hartley, and Weir (1994), and Bishop et al. (2000). The composition of the child's conversation was characterised by the proportion of utterances coded as initiations, maintenance or response behaviours relative to the total number of utterances of the child. The coding of all utterances provided insight into discourse management and the following-in behaviour of a partner's turn (see Table 3 for details). After coding all utterances, discourse participation and assertiveness indices were computed as suggested by Adams and Lloyd (2005, Table 3).

INSERT TABLE 3

The Targeted Observation of Pragmatics in Children's Conversation Observation scheme (TOPICC; Adams, Gaile, Freed, & Lockton, 2010) was used to obtain an *overall rating of the conversational competence* of the child, including taking account of listener's knowledge, discourse management and conversational responsiveness. The TOPICC scheme rates six categories or domains of pragmatic skills on a four-point scale: (1) reciprocity and turn taking, (2) taking account of listener's knowledge, (3) verbosity, (4) topic management, (5) discourse style, and (6) response behaviour (0, 'never' to 3, 'very frequent/always'). An example of a rated case is provided in Appendix 2. The ratings of

the items of each category were averaged which resulted in domain scores between zero and three. A domain score of 0 up to 1 was considered as unimpaired pragmatic skills. A domain score between 1 and 2 was considered to be mildly impaired, having only a slight impact on the interaction. A domain score higher than 2 was interpreted as severely impaired with a clear impact on the interaction.

Reliability. The third author (IB) was trained by the primary coder (EVDH) before assessing reliability. During the coding training, samples of six individuals (one of each syndrome group, two of each control group) who did not participate in the follow-up study (Time 2) were transcribed in order to obtain a transcript and coding agreement of at least 80%. When this level of agreement was reached, conversation samples of ten cases (30.30% of the total samples, two of each syndrome group, three of each control group) were randomly selected and coded. Intra-class correlations (ICC; Hallgren, 2012) were used for assessing inter-rater reliability, examining level of similarity between the two authors using the coding definitions presented in Table 3. The resulting average-measure intra-class correlation coefficients within a two-way mixed effects model, accounting for coder x subject interactions are summarised in Table 3.

Guidelines of Cicchetti (1994) were used for interpretation of the 95% confidence interval (CI) of these ICC values. When the reliability coefficients are below .40, the level of clinical significance is poor. When coefficients are between .40 and .59, the level of clinical reliability is fair. The level of clinical reliability is good when coefficients are between .60 and .74. Values between .75 and 1.00 are considered to be excellent (Cicchetti, 1994). The participation and assertiveness indices fell within the excellent

range of agreement according to these guidelines and were used for further analysis. The ICC for TOPICC scores was .91, 95% CI [.42, .98].

Statistical analysis

Although groups were well matched for CA and NVMA, our matched design did not allow us to control for all confounding factors that might contribute to differences across groups. Therefore, independent statistical procedures were preferred over paired statistics (Pearce, 2016). Kruskal-Wallis tests and post-hoc Mann-Whitney *U* tests for group comparisons were applied at Time 1 since the assumptions for normality and homogeneity of variance of several outcome variables were violated. We calculated effect sizes using the formula $r = z/\sqrt{n}$ (Field, 2013). Correction for multiple testing was applied using False Discovery Rate (FDR) control (Benjamini & Hochberg, 1995).

To compare developmental changes across groups, difference scores (Time 2 outcomes minus Time 1 outcomes) were computed. These difference scores (DIFF) indicate the degree and direction of changes in conversational behaviours between the two time points. Since not all difference scores met the assumptions for parametric analysis (Shapiro-Wilk <.050) and because of the limited number of participants, non-parametric statistical tests were used to analyse differences in developmental changes across groups. False Discovery Rate control (Benjamini & Hochberg, 1995) was used to correct for multiple comparisons.

Results

We analysed discourse management and following-in on a partner's turn by means of (1) proportions of maintenance utterances and responses, and (2) the participation and

assertiveness indices as described by Bishop et al. (2000). The TOPICC global and subscale scores at Time 1 shed further light on overall conversational competence, including the ability to take account of listener's knowledge, discourse management and response behaviour. We compared the performance of children with 22q11.2DS to the performance of children with IID and children with IID+ASD. Next, similar comparisons were made for the Williams syndrome group. Secondly, developmental changes (i.e. Time 2 minus Time 1, difference scores) in utterances and responses, assertiveness and TOPICC scores were described. Finally, we elaborated on differences between children with 22q11.2DS and children with WS.

Comparisons of conversational behaviour at Time 1

A. Children with 22q11.2DS compared to children with IID and children with IID+ASD

Discourse management, evaluated by means of **proportions of maintenance** utterances ($H(2) = 0.6$, $p = .932$) **and responses** ($H(2) = 1.09$, $p = .597$), was not significantly different across groups at Time 1. However, Kruskal-Wallis tests demonstrated a significant difference in the type of elaborations. Proportions of follow-up (F) statements ($H(2) = 6.69$, $p = .031$) were found to be significantly different across groups. Children with 22q11.2DS used significantly more follow-up statements (*Mdn* proportion F = .18, *range*: .09–.39) than children with IID (*Mdn* proportion F = .11, *range*: .04–.19); ($U = 9.50$, $p = .016$, $r = -.59$). Children with IID were more likely to use continuations to maintain the conversational topic going rather than follow-up statements (Table 4).

Conversational indices provided more insight into differences in discourse management across groups (Table 4). **Participation indices** ($H(2) = 6.07$, $p = .043$) and assertiveness indices ($H(2) = 6.36$, $p = .037$) were significantly different across groups at

Time 1. Mann Whitney *U* tests indicated no significant differences in the participation index between children with 22q11.2DS and children with IID (U = 15.00, p = .074). However, a significant difference in participation index was found between children with 22q11.2DS and children with IID+ASD (U = 10.00, p = .021). After applying FDR control this between-group difference could no longer be considered significant. It should be noted that in the 22q11.2DS group none of the children had a participation index (Table 4) higher than one, indicating that these children were not likely to take a dominant role in the conversation.

INSERT TABLE 4

Post hoc Mann-Whitney *U* tests indicated differences in the **assertiveness indices** of the 22q11.2DS group and the IID+ASD group (U = 10.00, p = .020) and of the IID and IID+ASD group (U = 11.50, p = .027). Although there was a tendency for children with IID+ASD to be more assertive due to consistently initiating new topics within their own interests, the between-group differences became non-significant after FDR control for multiple testing.

The **global TOPICC scores** were significantly different across groups ($H(2)$ = 14.60, p = .001). The median TOPICC score of children with 22q11.2DS was 17 (*range* = 13–19, *interquartile range* = 16–18) and was significantly higher than in children with IID (*Mdn* = 14, *range* = 6–16, *interquartile range* = 11 – 15), (U = 5.50, p = .004, r = -.70). The children with IID+ASD (*Mdn* = 22, *range* = 16–28, *interquartile range* = 17–25) had significantly higher TOPICC scores than the children with IID (U = 0.00, p < .001, r = -.83). The TOPICC scores of children with 22q11.2DS were not found to be significantly different from those of children with IID+ASD (U = 15.50, p = .088).

Subscale score analyses (Table 5) indicated that groups were rated differently for turn taking ($H(2) = 6.24$, $p = .043$), taking account of listener's knowledge ($H(2) = 17.39$, $p < .001$), topic management ($H(2) = 7.72$, $p = .016$), and discourse style ($H(2) = 8.79$, $p = .010$). Post hoc comparisons indicated that subscale scores for turn-taking ($U = 11.00$, $p = .029$) and for discourse style were significantly higher in children with 22q11.2DS compared to children with IID ($U = 9.00$, $p = .011$, $r = -.63$). The difference in discourse style remained significant after FDR control, however the difference in turn-taking scores did not. Children with 22q11.2DS were significantly less impaired in their ability to take account of listener's knowledge than children with IID+ASD ($U = 0.00$, $p = <.001$, $r = -.88$). Children with IID+ASD had significantly more difficulties with taking account of listener's knowledge ($U = 0.00$, $p = <.001$, $r = -.88$), discourse style ($U = 9.00$, $p = .014$, $r = -.62$) and topic management ($U = 9.00$, $p = .014$, $r = -.62$) than children with IID.

INSERT TABLE 5

B. Children with WS compared to children with IID and children with IID+ASD

The **proportion of maintenance utterances** ($H(2) = 0.54$, $p = .778$) and the **proportion of responses** ($H(2) = 1.94$, $p = .402$) did not differ across groups (Table 6). Kruskal-Wallis tests revealed significant differences for **assertiveness indices** (Table 6) across groups ($H(2) = 6.05$, $p = .49$), but not for the **participation indices** ($H(2) = 3.55$, $p = .17$). Children with WS started a new conversational turn more often than children with IID, causing a significant difference in assertiveness indices between-groups ($U = 10.00$, $p = .016$, $r = -.59$).

The global TOPICC scores were significantly different across groups ($H(2) = 15.56$, $p < .001$). The median TOPICC score of children with WS was 22 (*range* = 17–26,

interquartile range = 19–25) and significantly higher than in children with IID (*Mdn* = 14, *range* = 12–16, *interquartile range* = 14–15), (U = 0.00, p < .001, r = -.85). TOPICC scores of the IID+ASD group (*Mdn* = 22, *range* = 18–28, *interquartile range* = 20–25) were very similar to those of children with WS and significantly higher than in children with IID (U= 0.00, p < .001, r = -.85).

When analysing the **six subscales of TOPICC**, significant between-group differences were demonstrated for taking account of listener's knowledge (H(2) = 5.03, p = .021), topic management (H(2) = 8.85, p = .006) and discourse style (H(2) = 10.45, p = .002). Post hoc analyses showed that children with WS had significantly more difficulties with topic management (U = 9.00, p = .014, r = -.61) and a more impaired discourse style (U = 6.00, p = .005, r = -.69) than children with IID. Children with WS seemed to have fewer problems with taking account of listener's knowledge than children with IID+ASD (U = 11.50, p =.024). However, this result was considered non-significant after FDR control. Children with IID and IID+ASD were rated as significantly different in taking account of the listener's knowledge (U = 10.50, p = .030, r = -.59), topic management (U = 7.50, p = .007, r = -.65) and discourse style (U = 9.00, p = .016, r = -.63).

****INSERT TABLE 7****

Developmental changes

A. Children with 22q11.2DS compared to children with IID and children with IID+ASD

Despite the limited differences in discourse management at Time 1, several between-group differences became more pronounced over time. The difference scores (Time 2 minus Time 1) for **proportion of responses** (H(2) = 6.65, p = .032) were found to be significantly different across groups (Table 4). Post hoc Mann Whitney U analysis

showed that the course for the proportion of responses was significantly different between the 22q11.2DS and IID group (U = 10.00, p = .017, r = -.58). Seemingly opposite courses were observed across groups. We found an increase in the proportion of responses in four children with 22q11.2DS ($M_{increase}$ = 0.12) and a stable proportion in the other half of the group. In children with IID we noted different and mixed developmental courses. Six children with IID showed a decrease in the proportion of responses ($M_{decrease}$ = -0.10). One child had a stable proportion of responses and one an increased proportion of 0.13. Five children with IID+ASD showed an increase in proportion of responses ($M_{increase}$ = 0.13), one child showed a decrease of -0.18 and two children showed a stable proportion of responses.

Significant differences in developmental changes for **assertiveness indices** were found across groups (H(2) = 9.19, p = .004). Post Hoc analyses revealed significantly different developmental changes for *assertiveness indices* between the 22q11.2DS and IID group (U = 6.50, p = .005, r = -.67). No difference in the course of the **participation indices** could be demonstrated (H(2) = 4.73, p = .092).

In children with 22q11.2DS the **TOPICC global score** decreased over time in half of the group ($M_{decrease}$ = 4.75) and remained relatively stable (i.e. maximum change of +/- 2 points) in the other half. In the IID group the score remained relatively stable in all but one participant who showed a decrease of 4 points. In the IID+ASD group the TOPICC scores of three children remained relatively stable, in one child the TOPICC score increased 4 points over time and in four children TOPICC score decreased ($M_{decrease}$ = -6.25). The mixed profiles across groups prevented to demonstrate significant differences in developmental change of global TOPICC scores (H(2) = 2.99, p = .23).

Analysis of the **TOPICC subscales** revealed different developmental changes (Table 5) for taking account of listener's knowledge ($H(2) = 13.38$, $p < .001$) and discourse style ($H(2) = 6.57$, $p = .045$) across groups. Subsequent Mann-Whitney *U* analyses demonstrated a difference in the changes of discourse style ratings when comparing children with 22q11.2DS to children with IID ($U = 10.00$, $p = .018$). This result was considered as non-significant when applying FDR control. A difference was found for the change in taking account of listener's knowledge when comparing children with 22q11.2DS to children with IID+ASD ($U = 5.00$, $p = .004$). This result needed to be considered non-significant after applying FDR control. A statistically significant difference was demonstrated in the change scores for taking account of listener's knowledge when comparing children with IID and IID+ASD ($U = 2.00$, $p = .001$, $r = -.81$).

B. Children with WS compared to children with IID and children with IID+ASD

No significant differences were demonstrated in the course of the **proportions of maintenance utterances** ($H(2) = 4.22$, $p = .12$) or of the proportions of **responses** ($H(2) = 5.72$, $p = .06$) across groups. Consequently, no significant differences were found in the developmental changes of **participation** ($H(2) = 4.22$, $p = .12$) **or assertiveness indices** ($H(2) = 3.85$, $p = .16$) (see Table 6 for details).

Although no significant differences in the changes of the **global TOPICC scores** were found ($H(2) = 4.27$, $p = .12$), significant between-group differences in developmental change for **TOPICC subscales** of verbosity ($H(2) = 7.86$, $p = .016$) and taking account of listener's knowledge ($H(2) = 7.06$, $p = .025$) were demonstrated (see Table 7 for details). Post Hoc Mann Whitney *U* analysis demonstrated that in children

with WS the problems with taking account of listener's knowledge became more pronounced over time, whereas in children with IID+ASD the score remained stable or only slightly decreased resulting in a different developmental pattern for this domain (U = 10.50, p = .023). This was found to be non-significant after FDR control. No significant differences were found for the change in verbosity compared to the WS group.

Cross-syndrome differences

Our final aim was to compare the conversational behaviour of the two syndrome groups. At both time points no differences could be demonstrated in **proportions of maintenance utterances, responses, participation and assertiveness indices**. However, **the global TOPICC score** was significantly higher in children with WS than in children with 22q11.2DS (U = 5.00, p =.003, r = -.72). Children with WS had significantly higher ratings for the discourse topic management domain (U = 5.00, p = .002, r = -.72) at Time 1. No differences in the rate of change were found when comparing both microdeletion syndrome groups.

Discussion

Overall, this study has provided some evidence that children with specific microdeletion syndromes show atypical patterns of conversational ability compared to controls, some of which are syndrome specific.

Conversational characteristics of children with 22q11.2 deletion syndrome

In our sample, children with 22q11.2DS took a less dominating role in a conversation than children with IID and children with IID+ASD. They exhibited difficulties with active initiating, resulting in a lower assertiveness index compared to children with IID

and IID+ASD. However, this result did not survive correction for multiple testing. Children with 22q11.2DS used significantly more follow-up statements, i.e. additional optional contributions that did not elicit or provide information, than children with IID. This finding suggests that children with 22q11.2DS contribute less to topic maintenance. The global TOPICC scores of children with 22q11.2DS were significantly higher than those of children with IID, caused by a distant discourse style. Our findings at Time 1 therefore endorse the suggestions of Golding-Kushner et al. (1985) on withdrawn and less responsive behaviour in young children with 22q11.2DS and the initiating problems in primary school-aged children with 22q11.2DS reported by Persson et al. (2006) and Van Den Heuvel et al. (2016).

Over the last few years there has been a lot of controversy over the presence or absence of ASD features in children with 22q11.2DS (e.g. Angkustsiri et al., 2014; Niklasson et al., 2009). Van Den Heuvel et al. (2017) showed some similarities and differences in the socio-communicative behaviour reported by parents of children with 22q11.2DS and parents of children with IID+ASD. The present study also provides evidence for overlap and differences in conversational profiles of these groups. Children with 22q11.2DS differed from children with IID+ASD in terms of taking listener's knowledge into account. Children with IID+ASD were also more likely to talk at cross purposes and failed to establish an adequate communicative interaction resulting in a significantly higher TOPICC score compared to peers with IID and 22q11.2DS. These findings might be linked to greater social-cognitive abilities in children with 22q11.2DS or different mechanisms underlying the social cognitive deficits in both groups as suggested by McCabe et al. (2013).

Until now, no information was available regarding developmental changes in conversational competences of children with different neurodevelopmental disorders. This study revealed that children with 22q11.2DS showed less social engagement over time and frequently needed to be pushed for an answer at Time 2. In comparison to children with IID, children with 22q11.2DS seemed to have increasing difficulties with initiating and carrying on extended conversational sequences, resulting in significantly different developmental patterns for assertiveness and proportion of responses. However, the changes in TOPICC scores did not differ across groups. In our 22q11.2DS group, considerable variation was noticeable at Time 1 but this was less pronounced in comparison to children with IID+ASD in the follow-up phase. This might suggest that the communicative profile of children with 22q11.2DS becomes more uniform (and their role more passive) as they grow into adolescence. We hypothesise, in agreement with Bishop et al. (2000), that due to the limited verbal contributions of children with 22q11.2DS at Time 2, pragmatic shortcomings are simply more likely to be overlooked.

Overall, we observed that children with 22q11.2DS become less 'active' conversationalists over time. This is in contrast to the findings in typically developing children who become more dominating and involved in the conversation from primary school-age onwards (e.g. Baines & Howe, 2010, McTear & Conti-Ramsden, 1992; Short-Meyerson & Abbeduto, 1997).

Conversational characteristics of children with Williams syndrome

In line with previous studies (e.g. Skwerer et al., 2011; Stojanovik et al., 2001), children with WS had a relatively uninhibited conversational style and tended to dominate the conversation. Given their sensitivity to the social aspects of the conversational situation,

they produced more initiations resulting in significantly higher assertiveness and higher global TOPICC scores than children with IID.

When analysing the TOPICC global and subscale scores for children with WS, the key difference from other groups is that they have greater difficulties with discourse topic management. Despite their impressive talkativeness they could not conceal their lack of topic management strategies. An overload of details beyond the context requirements forced the listener to consistently infer the intended core message. Furthermore, favoured topics tended to recur, often those related to intense emotions (e.g. hospital visits, family gatherings) or activities with intensified sound levels (e.g. riding a tractor, driving a motor cycle, travelling by train). These behaviours led to significantly higher ratings on the topic management and discourse style domains of TOPICC compared to children with IID.

In terms of global TOPICC scores, children with ID+ASD and children with WS appeared to have equivalent levels of pragmatic language impairment. However, children with ID+ASD and WS had high global TOPICC scores for very different reasons. The key difference between children with ASD and the other groups of children in this study is that they are more impaired on the TOPICC subscale 'taking account of listener's knowledge'.

Although some similarities were evident between the profile of children with WS and that of children with IID+ASD, some differences should also be highlighted. First, the within-group variability in children with IID+ASD was larger than in children with WS. This corroborates the findings of Noens and van Berckelaer-Onnes (2005) that despite having good basic language abilities, children with ASD may encounter divergent

challenges during complex, advanced interactions such as conversations. These limitations are likely to be caused by a lack of social awareness and poor sensitivity to contextual cues that other children use to facilitate understanding of conversational contributions. Therefore, the range of results in children with IID+ASD was found to be very wide, highlighting a broad continuum varying from reluctant behaviour to excessively unconventional verbal behaviour. Children with IID+ASD were also more likely to not respond to questions at all or to ignore the initiations of the examiner, which seldom occurred in children with WS.

Interestingly, the children with WS seemed to get worse at taking listener's knowledge into account between Time 1 and Time 2, whereas children with ASD did not. However, it should be pointed out that at Time 2 the nonverbal mental age of children with WS was no longer perfectly matched to that of the IID+ASD group.

Finally, we would like to address a notable observation after analysing the comments on the TOPICC scheme and conversation transcriptions. In the majority of children with WS a lack of semantic specificity and word retrieval problems were observed. Incomplete sentences, reformulations and apparent disfluencies might indicate that children with WS encountered difficulties with monitoring their output and message organisation. They often interrupted the examiner to re-start (unfinished) sentences or story lines, which contributed to confusion. We suggest that these difficulties may have influenced the rating of the topic management domain. Overall, parents and caregivers should be aware that the persisting difficulties with interpreting contextual cues and perseverative talk about specific topics make children with WS vulnerable for communicative breakdowns and social rejection (Riby et al., 2014). This may place them

in a downward spiral that might partly explain the changes in conversational behaviours over time.

Quantitative analyses versus TOPICC scores

Although we did not perform a correlation analysis between conversational turns and TOPICC scores, our in-depth analyses elucidated some similarities and differences between the two measures in their ability to characterise conversational proficiency across groups.

The TOPICC scheme allowed us to differentiate children with microdeletion syndrome from children with IID. Moreover, TOPICC subscale ratings allowed us to illustrate clear differences between conversational behaviours of children with IID and children with IID with comorbid ASD. Therefore, our exploratory findings suggest that the TOPICC scheme might have the potential to demonstrate pragmatic shortcomings in diverse groups of children with neurodevelopmental disorders. Developmental changes in conversational competence over time could not be deduced from the global TOPICC score but some subscales ratings changed differently across groups. Since we only assessed the children at two time points with a fairly short interval of 18 to 24 months, this finding should be interpreted with caution. Further research is needed to analyse if TOPICC ratings are sensitive to subtle developmental changes.

Limitations and future directions

Matching diverse groups of children with ID is challenging and several procedures have been criticised (e.g. Jarrold & Brock, 2004; Mervis, 2004). This study applied a group matching procedure controlling for chronological age and nonverbal mental age. Although we tried to reduce the impact of cognitive differences and age benefits, some

differences across groups for structural language ability may have impacted the conversational outcomes. Several other individual characteristics such as medical factors, neuropsychological factors (e.g., executive functions, working memory), socioeconomic status, and environmental factors (e.g. content and frequency of communicative intervention) may have influenced the results and affected developmental change (Bishop et al., 2000; Swillen, 2016). Moreover, exact matching of all core skills wass not feasible. Therefore, the impact of these factors on pragmatic language development should be further explored in future research.

Small sample sizes are common in labour-intensive longitudinal studies of conversational competences but often lack the power to find clear-cut differences across groups. The considerable variation in all groups also prevents us from generalising our findings to these wider populations. The use of FDR control to account for Type I errors prevented us from demonstrating some potentially significant differences across groups. This control for multiple testing might have increased the probability for Type II errors. Hence, the results of the present study are preliminary and large-scale longitudinal studies with regular follow-ups throughout the lifespan are needed. It is also important to bear in mind that significant changes might have arisen simply as a result of some typical variations in the unstructured assessment procedures or narrow contextual sampling (Adams, Green, Gilchrist, & Cox, 2002; Adams & Lloyd, 2005). However, the current findings confirm clinical observations and parental reports and contribute to the delineation of socio-communicative courses in children with 22q11.2DS and WS.

Conclusions and clinical implications

Overall this study highlights that children with different genetic and neurodevelopmental disorders have different strategies for continuing a conversation and different awareness for social cues and listeners' communicative needs. Our longitudinal findings suggest that over time children with IID become more actively involved in the conversation, indicated by a higher proportion of maintenance behaviour and a lower proportion of responses. This is in contrast to children with 22q11.2DS who tended to have an increasingly more passive conversation style over time, indicated by a different developmental course of assertiveness and responses in comparison to children with IID. Children with WS differed from both children with IID and children with 22q11.2DS in their competence at managing the topic structure of a conversation.

In children with 22q11.2DS and WS, appropriate as well as inappropriate conversational variants were observed. Some of their conversational behaviours resembled the profile of children with IID+ASD. However, several results suggest more severe conversational problems in children with IID+ASD related to a profound deficit in the ability to take account of listener's knowledge. Therefore, it would seem valuable to explore differences in social cognitive abilities across groups that may underlie these differences in conversational competence. Children with 22q11.2DS and WS can be seen as socially more incompetent, and are prone to be less preferred as conversational partners (Hemphill & Siperstein, 1990). Therefore, remediation of conversational skills and support of socio-cognitive discourse awareness may foster advances in everyday communicative interactions and result in a more positive peer response. Specifically, active initiating and avoidance of inappropriate pauses appear to be areas that deserve emphasis in conversational skills training in children with 22q11.2DS. In children with

WS help understanding of the conversational balance and finding common ground may improve information transfer. Given the divergent courses and persisting conversational shortcomings, replication of this research and further follow-up are both desirable. This may lead us towards anticipatory, individually tailored socio-communicative goals that can be targeted in school or at home with the ultimate aim of optimising quality of life in these groups of children.

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TABLE TITLES

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Table 2. Group characteristics of the WS group and IID and IID+ASD control groups.

Table 3. Overview of codes and indices for quantitative analysis of conversational turns and intra-class correlations coefficients with 95% confidence interval (CI) for inter-rater reliability.

Table 4. Total number of utterances, proportion of maintenance and response utterances, and conversational indices at Time 1 and developmental changes in children with 22q11.2DS and IID and IID+ASD controls

Table 5. TOPICC subdomains scores (0-1 = unimpaired; 1-2 mildly impaired with slight impact on the interaction; ≥ 2 severely impaired with clear impact on the interaction) at

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Table 1. Group characteristics of the 22q11.2DS group and IID and IID+ASD control group

	22q11.2DS (*n* = 8) 4 male, 4 female			IID (*n* = 8) 7 male, 1 female			IID+ASD (*n* = 8) 6 male, 2 female			Exact *p*° 22q–IID		Exact *p*° 22q–IID+ASD	
	Time 1	Time 2	Diff.	Time 1	Time 2	Diff.	Time 1	Time 2	Diff.	T1	T2	T1	T2
CA [a]													
Mdn	9.6	11.4	1.10	9.8	11.4	1.9	9.11	11.7	1.7	.902	.857	.984	.942
Range	7.7–13.1	9.1–15.1	1.6–2.0	7.11–12.5	9.5–14.5	1.6–2.0	6.5–12.5	7.11–14.1	1.6–1.9				
NVMA[b]													
Mdn	6.8	8.0	0.9	7.0	7.2	0.1	6.6	7.9	1.6	.999	.137	.562	.747
Range	6.2–9.1	6.2–9.7	0–3.0	6.2–7.8	6.3–7.11	-0.7–0.9	6.0–7.5	6.8–11.0	0.1–3.7				
CLS[c]													
Mdn	67	72	2.5	63	60	-3.5	66	69	0.0	.267	.023*	.903	.775
Range	55–80	55–80	-10–10	57–72	57–71	-10–13	57–77	55–87	-7–17				
SCS[d]													
Mdn	3.5	4.5	1.0	2.5	6.5	0.5	4.5	5.0	1.5	.628	.639	.737	.627
Range	1–12	1–10	-3–7	1–7	1–11	-2–9	1–10	2–8	-2–2				
FSS[e]													
Mdn	4.5	5.5	1.0	3.0	2.0	0.0	3.0	5.0	0.5	.418	.045*	.564	.852
Range	1–8	1–9	-4–5	1–6	1–6	-4–0	1–7	1–7	0–5				

Notes.
[a] CA = chronological age (years.months), matching variable
[b] NVMA = nonverbal mental age (years.months), averaged age equivalents of nonverbal reasoning subtests (G*f* index), matching variable
[c] CLS = core language score ($X \sim N$ (100,15)), composite of CELF–P2–NL or CELF–4–NL subtests reflecting overall structural language level
[d] SCS = sentence comprehension score ($X \sim N$ (10,3)), scaled scores of Sentence Structure subtest CELF–P2–NL or Concepts and Following Directions subtest CELF–4–NL, reflecting the sentence comprehension level
[e] FSS = formulating sentences score ($X \sim N$ (10,3)), scaled scores of Formulating Sentences subtest CELF-4-NL, reflecting sentence production level
° Mann-Whitney *U* test for independent samples, if $p > .500$ groups can be considered equivalent on the matching variable (Frick, 1995)
*p <.050, significant group difference at Time 2; Diff. = difference score (Time 2 minus Time 1).

Table 2. Group characteristics of the WS group and IID and IID+ASD control groups.

	WS (*n* = 8) 5 male, 3 female			IID (*n* = 8) 6 male, 2 female			IID+ASD (*n* = 8) 7 male, 1 female			Exact *p*° WS–IID		Exact *p*° WS–IID+ASD	
	Time 1	Time 2	Diff.	Time 1	Time 2	Diff.	Time 1	Time 2	Diff.	T1	T2	T1	T2
CA [a]													
Mdn	8.6	10.4	1.10	8.7	10.3	1.9	8.11	10.7	1.8	.722	.821	.942	.874
Range	6.4–12.5	7.10–14.5	1.6–2.0	7.1–12.8	8.7–14.6	1.6–1.11	6.5–12.5	7.11–14.1	1.6–2.0				
NVMA[b]													
Mdn	6.5	6.11	0.11	6.4	6.10	0.4	6.0	7.1	1.5	.817	.520	.999	.313
Range	5.0–8.7.1	5.9–8.2	-2.4–2.4	5.4–8.8	5.10–7.5	-1.4–1.10	4.8–8.11	6.5–10.3	0.5–2.8				
CLS[c]													
Mdn	65	67	0	60	66	1.5	64	72	5	.699	.943	.905	.290
Range	55–85	55–79	-13–5	55–71	55–80	-4–13	55–77	55–87	-5–14				
SCS[d]													
Mdn	4.5	4	-1	2.5	5.0	2.5	3.5	4	0.5	.693	.083	.727	.418
Range	1–6	1–4	-2–2	1–7	1–11	-2–9	1–10	1–8	-2–2				
FSS[e]													
Mdn	3.0	4.5	1	3.5	4.0	0	2.5	4.5	1	.968	.311	.777	.916
Range	1–8	1–7	-2–3	1–6	1–6	-2–1	1–7	1–7	0–3				

Notes.
[a] CA = chronological age (years.months)
[b] NVMA = nonverbal mental age (years.months), averaged age equivalents of nonverbal reasoning subtests (G*f* index)
[c] CLS = core language score ($X \sim N$ (100,15)), composite of CELF–P2–NL or CELF–4–NL subtests reflecting overall structural language level
[d] SCS = sentence comprehension score ($X \sim N$ (10,3)), scaled scores of Sentence Structure subtest CELF–P2–NL or Concepts and Following Directions subtest CELF–4–NL, reflecting the sentence comprehension level
[e] FSS = formulating sentences score ($X \sim N$ (10,3)), scaled scores of Formulating Sentences subtest CELF-4-NL, reflecting sentence production level
° Wilcoxon Signed Rank test for dependent samples, if $p > .500$ groups can be considered equivalent on the matching variable (Frick, 1995)
*p <.050, significant group difference at Time 2; Diff. = difference score (Time 2 minus Time 1).

Table 3. Overview of codes and indices for quantitative analysis of conversational turns and intra-class correlations coefficients with 95% confidence interval (CI) for inter-rater reliability.

Coding category*	***Code***	***Definition***	***ICC***	***95% CI***
Maintenance utterances				
Continuation	C	Utterance which continues or elaborates on a previous utterance within a conversational turn.	.96	[.82, .99]
Follow-up	F	Additional optional contribution, which does not elicit or provide information. It encourages the interlocutor to continue the conversation and acknowledges a given response.	.99	[.97, .99] .
Response behaviour			.99	[.95, .99]
Conversational indices				
Participation index		Ratio of C utterances (Total N of C utterances in 100 conversational turns) to E utterances (Total N of E utterances in 100 conversational turns). An index > 1.0 suggests the child dominated the conversation (Lloyd, Lieven, & Arnold, 2001).	.99	[.97, .99]
Assertiveness index		*Total N of Utterance C divided by Total N of Utterance E* The child's tendency to start a new conversational turn was measured by dividing the first parts (IS+IQ) by the total number of child's utterances in 100 conversational turns. *Child's N of IQ + IS divided by Total N of Utterances C*	.96	[.82, .99]

Notes.
*Selection and adaption of conversational parameters suggested by Adams and Bishop (1989), McTear (1985), Adams & Lloyd (2005), and Bishop et al. (1994, 2000), only parameters with excellent agreement were included for further analysis.
C = Child; E = Examiner; N = Number; IS = Initiating statement; RC = Request for clarification

Table 4. Total number of utterances, proportion of maintenance and response utterances, and conversational indices at Time 1 and developmental changes in children with 22q11.2DS and IID and IID+ASD controls.

	Total N of utt.		**Prop. maintenance**		**Prop. responses**		**Participation index**		**Assertiveness index**	
	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.
22q11.2DS										
Mdn.	106	6.5	.45	.04	.44	.06	.72	-.11	.10	-.06
range	71–124	-13–23	.27–.75	-.18–.06	.31–.54	-.02–.23	.54 – .85	-.23 – .10	.06 – .19	-.09 –.03
IQR	100–118	-9.5–11.5	.34–.50	-.01–.05	.32–.54	-.02–.10	.70 – .81	-.17 – -.03	.07 – .17	-.09 – -.03
IID										
Mdn.	107.50	19.50	.37	.14	.54	-.12	.89	.03	.10	.01
range	85–158	-23–43	.17–.67	-.04–.29	.27–.75	-.30–.13	.48 – 1.06	-.33 – .20	.03 – .23	-.07 – .04
IQR	90.25–145.25	3.25–21	.23–.57	.03–.22	.27–.67	-.22–.03	.68 – .99	-.16 – .13	.07 – .15	-.05 – .03
IID+ASD										
Mdn.	119.50	-11	.35	.08	.51	.05	.92	-.15	.16	-.11
range	112–146	-19–14	.23–.58	-.13–.28	.33–.63	-.25–.20	.62 – 1.10	-.36 – -.03	.10 – .23	-.07 – .04
IQR	112.75–129	-13.75–10.5	.30–.50	-.05–.15	.33–.52	-.15–.13	.79 – 1.07	-.28 – -.06	.15 – .21	-.15 – -.05

Notes.
N = number; utt. = utterances; IQR = interquartile range; Diff. = difference score (Time 2 minus Time 1)

Table 5. TOPICC subdomains scores (0-1 = unimpaired; 1-2 mildly impaired with slight impact on the interaction; ≥ 2 severely impaired with clear impact on the interaction) at Time 1 and developmental changes in children with 22q11.2DS, IID and IID+ASD controls

	Turn-taking		**Taking account of listener knowledge**		**Verbosity**		**Topic management**		**Discourse style**	
	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.
22q11.2DS										
Mdn.	2	-0.17	1.50	0	0.25	-0.25	1	0	1	-0.34
range	1.33–2.67	-1–0.33	0.50–1.50	-0.50–1.50	0–2	-2–0	0.67–1.67	-0.67–0.67	0.33–1.00	-0.67–0.34
IQR	1.42–2.33	-0.58–0	1.13–1.50	0–0.50	0–1.46	-1.25–0	1–1.25	-0.59–0.25	0.42–1.00	-0.67–0.08
IID										
Mdn.	1.33	-0.33	1.50	0.25	0.50	0	0.67	0	0.33	0.34
range	0.67–1.67	-1–0.66	0.50–1.50	0–0.50	0–1.50	-0.67–0.50	0–1.50	-0.83–0.66	0–0.67	-0.34–0.34
IQR	1–1.67	-0.58–0	1.00–1.50	0–0.50	0–0.92	0–0.38	0.67–1.25	-0.34–0.58	0.08–0.59	0.08–0.34
IID+ASD										
Mdn.	1.67	0	2.00	-0.50	1	-0.50	1.67	0	1	0
range	1–2	-0.67–0.34	2–3	-1–0	0–2.50	-1.50–0.50	0.67–2.67	-1–1	0.33–2.00	-0.67–1
IQR	1.33–1.67	-0.33–0.25	2–2.38	-1–-0.50	0–1.88	-1–0	1.08–2.25	-0.67–0.58	0.42–1.59	-0.33–0.33

Table 6. Total number of utterances, proportion of maintenance and response utterances, and conversational indices at Time 1 and developmental changes in children with WS and IID and IID+ASD controls.

	Total N of utt.		**Prop. of maintenance**		**Prop. of responses**		**Participation index**		**Assertiveness index**	
	Time 1	Diff**.**	*Time 1*	Diff.	*Time 1*	Diff.	*Time 1*	Diff.	*Time 1*	Diff.
WS									.	
Mdn.	116.50	-12.00	.38	.01	.46	.01	.83	-.14	.17	-.02
range	101–141	-23–12	.24–.58	-.05–.21	.23–.54	-.14–.09	.68 – 1.06	-.34 – .08	.09 – .19	-.16 – .06
IQR	109.75–119.75	-18.25– -2.75	.26–.46	-.05–.08	.37–.53	-.08–.07	.72 – .93	-.25 – .02	.15 – .19	-.09 – .04
IID										
Mdn.	103.50	11.00	.32	.14	.56	-.13	.69	.08	.09	-.01-.05
range	83–149	-6–70	.17–.67	.01–.29	.26–.78	-.24–.01	.53 – 1.07	-.24 – .48	.04 – .17	– .04
IQR	92.25–131.50	0.5–20.5	.21–.51	.03–.20	.33–.72	-.21– -.06	.63 – .96	-.16 – 23	.07 – .15	-.03 – .03
IID+ASD										
Mdn.	115	-1	.34	.08	.47	.01	.95	-.14	.16	-.08
range	84–146	-19–27	.23–.52	-.13–.28	.27–.75	-.25–.20	.64 – 1.21	-.55 – .13	.02 – .23	-.15 – .10
IQR	103–131	-12.25–22.25	.30–.50	.01–.15	.33–.59	-.17–.13	.80 – 1.09	-.31 – .02	.10 – .22	-.14 – -.04

Notes.
N = number; utt. = utterances; IQR = interquartile range; Diff. = difference score (Time 2 minus Time 1)

Table 7. TOPICC subdomains scores (0-1 = unimpaired; 1-2 mildly impaired with slight impact on the interaction; ≥ 2 severely impaired with clear impact on the interaction) at Time 1 and developmental changes in children with WS, IID and IID+ASD controls

	Turn-taking		**Taking account of listener knowledge**		**Verbosity**		**Topic management**		**Discourse style**	
	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.	Time 1	Diff.
WS										
Mdn.	1.67	-0.17	2	0.25	1.25	-0.50	1.67	-0.33	1.67	0
range	1–2	-1–0.67	0.50–2.50	-1–1.50	0.50–2.50	-2–1	1.33–2.67	-1.50–0	0.33–1.00	-0.67–1
IQR	1–1.67	-0.67–0.59	1.00–1.50	0–0.50	0.63–1.88	-0.50–0	1.37–2.25	-0.92–0	0.42–1.00	-0.59–0.34
IID										
Mdn.	1.50	-0.33	1.25	0	0.50	0.25	1.00	0.33	0.33	0.34
range	0.67–2.33	-1.33–0	1–2	-1–0.67	0–1.50	-0.50–1.50	0.67–2.33	-0.66–0.67	0-0.67	-0.34–1
IQR	1–2.17	-0.92–-0.08	1–1.5	0–0.38	0–0.88	0–1.00	0.67–1.38	-0.46–0.68	0.08–0.67	0–0.58
IID+ASD										
Mdn.	1.50	0	2	-0.50	1.75	-0.75	1.84	-0.50	0.84	0
range	1–2.67	-0.67–1	1–3	-1.50–0	0–2.50	-2–0	1.33–2.67	-1–0.66	0.33–2.00	-1–0
IQR	1.33–1.67	-0.59–0.26	1.63–2.38	-1–0	0.13–2.38	-1.38–0	1.67–2.25	-0.67–0.25	0.67–1.50	-0.50–0.33

APPENDICES

Appendix 1. Content of elicitation pictures

	Theme, content and elicitation goals
Picture 1	Theme: Negative experience – falling/hurting oneself Pictured content: Boy who injured his knee and a woman putting a bandage on the wounded knee. A bicycle is lying on the ground suggesting that the boy fell off his bicycle. Elicitation goal: The aim of this picture was to stimulate the child to talk about a negative experience of falling or hurting himself and the consequences of this event.
Picture 2	Theme: Celebration – Birthday party Pictured content: One woman and two children were shown in the picture. A girl embraced the woman, suggesting she was celebrating her birthday. The boy in the picture looked at the birthday cake. Elicitation goal: The aim was to engage the child to talk about his own birthday and participating in birthday celebrations.
Picture 3	Theme: Playing outdoors Pictured content: Three children are in the garden throwing a ball to each other. Elicitation goal: The child was encouraged to talk about a game that he likes to play in- or outside the house. The child was also asked to outline one of his favourite games.

Appendix 2. Example of a TOPICC analysis (Adams et al., 2010) of a child with WS (Time 1)

	CHILD ID	Rater ID	Category rating (X)				Notes/Description of behaviour
	WS PAIR3	EVDH	*0*	*1*	*2*	*3*	
			never *typical mature behaviour*	***occasionally*** *slight impact on interaction*	***often*** *moderate to noticable impact on interaction*	***very frequent - always*** *abnormal behaviour marked impact on interaction*	
Category	**Reciprocity/ Turn-taking**						
1.1.	Difficulties responding to questions					X	a lot of false starts, clownish behaviour before response
1.2.	Interrupts speaker frequently, or frequent pauses				X		a lot of mazes (word finding problems?), interruptions
1.3.	Reticence		X				great willigness to interact
Category	**Taking account of listener knowledge**						
2.1.	Giving too much detail and information				X		details : colours, emotions
2.2.	Giving too little information					X	high number of request for clarification examiner
Category	**Verbosity**						
3.1.	Child dominates conversation			X			
3.2.	Child uses too many questions			X			asks frequently who? and how?
Category	**Topic management**						
4.1.	Obsessional Topics		X				
4.2.	Difficulties with topic maintenance			X			
4.3.	Stereotyped or unusual language				X		perseveration on hospital theme, talks about white things
Category	**Discourse style**						
5.1.	Proximity				X		short distance, touching examiner
5.2.	Overly formal or overly friendly					X	unnecessary, awkward laughs, extreme enthousiast
5.3.	Non verbal behaviours					X	uses non-verbal language too much, overload of gestures
Category	**Response problems**						
6.1	Comprehension or linguistic limitation results in odd responses					X	answer often doesn't matches the question, when child does not understand the questions she pauzes, looks around and she starts another topic, word finding problems?
		Ratio	2	3	4	5	
		Score per category	0	3	8	15	
		Total score (max. 42)	26				
		Severity percentage	61.90				