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Narrative language abilities in adults with Down syndrome: A remote online elicitation study using the Multilingual Assessment Instrument for Narratives (MAIN)

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Introduction: This research represents, to the best of our knowledge, the first attempt at assessing narrative retell remotely in people with Down syndrome and will provide valuable information on the validity and feasibility of remote online assessment with this population. Most research on language abilities in Down syndrome has focused on children and adolescents, making adults an understudied population. The present research seeks to establish a baseline of functioning for narrative language abilities in adults with Down syndrome, as part of a larger research aiming to investigate possible changes associated with aging and the emergence of Alzheimer's disease in this population.

Methods: We recruited 13 adolescents and young adults with Down syndrome aged 15–33 years (mean age: 21), matched to a control group of younger typically developing children aged 4–10 years (mean age: 6) on verbal Mental Age (MA). Participants completed a picture-based story retell activity from the Multilingual Assessment Instrument for Narratives (MAIN) and a series of standardized background measures of language and cognitive ability.

Results: Our analyses focused on macrostructural indices of narrative performance, narrative length and lexical diversity. Results revealed that our participants with Down syndrome were outperformed by verbal MA-matched controls on measures of story structure and story comprehension, as well as lexical diversity. No difference was found on total number of words, indicating the groups produced comparable amounts of speech despite differences in story grammar and lexis.

Discussion: We interpret the results in light of previous research on macrostructural narrative performance in adults and younger adolescents with Down syndrome. Recruitment and data collection outcomes are discussed in terms of successful strategies and possible improvements. We conclude that remote online assessment of people with Down syndrome is feasible, although considerations should be made with regards to facilitating enrolment, and task engagement.

Our participants demonstrated ability to engage with the experimenters over video chat and were able to complete the activities proposed mostly independently, with minimal involvement required from caregivers. Recommendations for future remote online studies involving children and people with intellectual disabilities are discussed.

KEYWORDS

Down syndrome, intellectual disability, narrative language, online remote elicitation, macrostructure

Introduction

Down syndrome is a developmental disorder associated with mild to moderate levels of intellectual disability (Chapman and Hesketh, 2000). In 95% of cases, the syndrome is caused by a full extra copy of chromosome 21 (trisomy), while in a small proportion of cases the extra copy is only present in some cells (mosaicism) or parts of chromosome 21 attach to another chromosome (translocation) (Martin et al., 2009). Down syndrome affects around 1 in 1,000 live births (Wu and Morris, 2013) and according to a 2015 estimate (de Graaf et al., 2021) over 41,000 people with Down syndrome live in the United Kingdom alone.

Over the past 2 years, the population with Down syndrome, amongst others, has been disproportionally affected by changes associated with the Covid-19 pandemic, particularly by the strict social-isolation regulations that have been enforced worldwide. Individuals with Down syndrome are prone to a range of physical and psychiatric complications associated with the syndrome which make them particularly vulnerable to the Covid-19 disease (Clift et al., 2021). In addition to more severe health risks associated with the SARS-CoV-2 virus, recent reports on the impact of the pandemic have evidenced an increase in depressive symptoms and worse overall functioning in adults with Down syndrome as a result of local lockdown measures (Villani et al., 2020). In light of the increased health risks associated with the current climate, research practices worldwide have had to adapt their methodology to minimize face-to-face contact with participants. As a result, many research teams have turned to remote approaches to language assessment. In the following sections, we review the cognitive and language profile of adults with Down syndrome, with a focus on aspects of narrative language abilities, and present the rationale for the present research. The study seeks to bring insights into the narrative language skills of adolescents and adults with Down syndrome and assess the feasibility of a remote online approach to recruitment and language elicitation in this population.

Cognitive and language abilities in Down syndrome

While a certain degree of heterogeneity must be recognized, individuals with Down syndrome often present with overall developmental delays, coupled with selective weaknesses in aspects of higher-order cognitive functioning. General patterns include later onset of language acquisition, impaired speech production, and impairments in working memory, especially phonological as opposed to visuospatial memory (Jarrold and Baddeley, 2001; Campbell et al., 2013). Both memory and language exhibit significant delays in this population relative to typically developing (TD) counterparts, however abilities in these domains are far from being homogeneously affected. Language, in particular, presents intriguing dissociations. Receptive language skills tend to be relatively stronger than production (Chapman et al., 1998; Miles and Chapman, 2002; Cleave et al., 2012), with vocabulary comprehension generally aligning with non-verbal mental age expectations (Abbeduto et al., 2003). Expressive language, on the other hand, is an area of marked difficulty: most individuals with Down syndrome produce shorter and more simplified sentences relative to what would be expected on the basis of non-verbal mental age (Chapman et al., 1998; Caselli et al., 2008; Price et al., 2008). Morphosyntax is particularly weak. Difficulties are reported in both comprehension and production of grammatical morphemes, with common errors involving omission or incorrect use of past tense -ed, third person singular -s, present progressive -ing, auxiliaries and articles (Fowler et al., 1994; Hesketh and Chapman, 1998; Eadie et al., 2002; Caselli et al., 2008). At the level of syntax, individuals with Down syndrome also show difficulties in the comprehension of complex sentences, such as passives, relative clauses, and interrogatives (e.g., Joffe and Varlokosta, 2007; Frizelle et al., 2019; Perovic and Wexler, 2019), as well as specific syntactic relations involved in the interpretation of reflexive pronouns (Perovic, 2006), across their lifespan. Less is known about the pragmatic skills of individuals with Down syndrome. Early studies report pragmatics to be a relative strength for both

children and adults with Down syndrome, especially compared to their grammatical skills (see Roberts et al., 2007, for a review). However, more recent investigations suggest that children with Down syndrome may perform poorer than younger controls in almost all areas of pragmatics, from topic initiation, topic elaboration and maintenance, to the use of context and conversational repairs (e.g., Smith et al., 2017).

Most of the available evidence on language abilities in Down syndrome comes from studies carried out with children and adolescents, making adults an understudied population. The relatively recent increase in life expectancy for people with Down syndrome (Strauss and Eyman, 1996) has invited interest into the cognitive changes that may be associated with aging in this population. Clinical studies have established a now well-researched association between Alzheimer's disease (AD) and the syndrome, indicating that people with Down syndrome are at ultra-high risk of developing the neurodegenerative condition (Sinai et al., 2012; Startin et al., 2019a). It is estimated that roughly 75% of individuals with Down syndrome aged over the age of 60 show clinical markers of Alzheimer's dementia (Lai and Williams, 1989, also see McCarron et al., 2014 and McCarron et al., 2017 for similar estimates), though clinical changes can be detected as early as 35 years of age (see Ballard et al., 2016). However, lack of a clear understanding of AD symptom progression in Down syndrome (see Lautarescu et al., 2017 for a review) brings the issue of (early) diagnosis in this population front and center. In fact, early detection of neurodegenerative conditions in populations with intellectual disability is greatly complicated by the presence of pre-existing lifelong cognitive impairments which make diagnosis difficult in the absence of previous baseline assessments (Devenny et al., 2000; Sinai et al., 2012). Few studies have focused on the extent to which language skills are affected by age- and dementia-related decline in the Down syndrome population, revealing inconsistent findings. For example, Devenny and Krinsky-McHale (1998) report no evidence of age-dependent language deterioration in adults with Down syndrome, while others have supported the idea of a decline in language skills from the fourth decade of life (e.g., Carter Young and Kramer, 1991; Cooper and Collacott, 1995; Perovic and Wexler, 2019). Nevertheless, a more recent publication argues that the inclusion of language assessment can be beneficial to improving the diagnosis of dementia in Down syndrome (Pulsifer et al., 2020). This suggests that monitoring language skills in adults with Down syndrome could generate useful insights into age- and AD-related changes in this population. In particular, honing in on language domains that could reveal language difficulties resulting from Alzheimer-related decline in adults without Down syndrome, such as narrative skills (e.g., Chapman et al., 1998; Ash et al., 2007), would provide valuable relevant information. Such information, in turn, would allow us to precisely map out the linguistic profile of the adult population with Down syndrome, and pinpoint the areas of language ability that could be problematic for those adults with Down syndrome at risk of developing Alzheimer's.

Narrative abilities in Down syndrome

One particularly fruitful method of assessing language production abilities of individuals with Down syndrome at both syntactic and pragmatic level relies on narrative tasks. Narratives are complex forms of discourse consisting of connected passages that primarily integrate information about events ("landscape of action") and participants' mental states ("landscape of consciousness," Bruner, 1986). Such productions, therefore, require careful coordination of interacting socio-cognitive and linguistic factors. On the one hand, well-structured narratives construct a hierarchical framework for the presentation of related events, while incorporating different levels of perspective from both the narrator and the participants in the story (Pearson and De Villiers, 2006). As such, storytelling ability is influenced by the speaker's prior life experiences and world knowledge, including their familiarity with themes and events in the story, their understanding of event sequences, temporal or causal relationships, and more general social and cultural dynamics (Miles and Chapman, 2002; Segal and Pesco, 2015). Being able to reason about others' mental states (the ability traditionally referred to as Theory of Mind, ToM) represents an important aspect of narrative competence, as it allows the narrator to make predictions and offer interpretations of characters' behaviors, as well as judgements about the listener's perspective and the common ground shared with the audience—all necessary to produce informative and relevant narratives (Matthews et al., 2018).

On the other hand, the expression of aspects relevant to event structure and participant's states will be influenced by the underlying linguistic competence of the speaker and their ability to formulate, through means of lexical choices and morphosyntactic organization, the linguistic scaffolding necessary for generating cohesive and comprehensible narratives. Evidence suggests, in fact, that pragmatic language skills such as the ones involved in storytelling are in large part directly related to formal language competence (Matthews et al., 2018), a finding which could have important consequences on how we interpret narrative performance. For example, a lack of references to character's mental states in narrative production could be indicative of ToM difficulties, but could also be explained in terms of an underlying syntactic impairment: mental state verbs often require more complex syntactic constructions (e.g., complement clauses) to be expressed, which could reveal problematic for young children and especially for individuals with language impairments (de Villiers and de Villiers, 2000 for an account of the role of language in the development of ToM). Production of narrative content, especially for individuals with intellectual disabilities, may

also be affected by sampling context: it has been evidenced that participants with DS tend to produce larger MLUs and more complex syntactic constructions in response to picture-supported narrative elicitation techniques as opposed to conversational samples, including pictureless narration (Miles and Sindberg, 2006). Narrative tasks can thus provide useful insights into a range of underlying linguistic, cognitive and social abilities of speakers, and have proven especially effective modes of language elicitation in populations with language and intellectual impairments (Norbury and Bishop, 2003). Particularly, in addition to providing a structured framework for the elicitation of speech samples (Sealey and Gilmore, 2008), narratives offer a way of evaluating production on two distinct levels of linguistic competence: macrostructure and microstructure. Macrostructure refers to higher order organizational and cohesive aspects of story structure, while microstructure refers to internal elements of linguistic constructions. Macrostructural analysis focuses on narrative competence from the perspective of hierarchical organization, by examining the presentation of story grammar elements and the structural complexity of story episodes. This might involve determining the number and structure of episodes contained in a narrative by identifying key elements such as initiating events, goals, and outcomes, as well as evaluating the use of narrative tools such as appendages, orientations and evaluations (Berman and Slobin, 1994). On the other hand, microstructural analysis focuses on evaluating the use and accuracy of morphosyntactic constructions, such as verb morphology, syntactic phrases and dependent clauses (Ukrainetz et al., 2005; Justice et al., 2006). In our brief review of studies examining narratives in adults with Down syndrome, we shall focus on macrostructure rather than microstructure. Microstructural skills are expected to be deficient across the lifespan, in view of the persistent grammatical deficits associated with Down syndrome. Macrostructural abilities, however, may show different patterns. As discussed above, pragmatics is generally considered an area of relative strength in individuals with DS. Since discourse cohesion and coherence are domains known to be affected by age-related decline in the population without Down syndrome (e.g., Ash et al., 2007), however, aspects of macrostructure may prove to be even more vulnerable in adults with Down syndrome who are approaching the age of dementia onset for this population.

Below we review a selection of studies that have examined macrostructural narrative language skills in individuals with Down syndrome. In light of the limited availability of studies retrieved focusing on *adult* language skills, we also include research carried out with older children and adolescents with Down syndrome. In line with previously discussed considerations regarding the effects of task structure and sampling context on the elicitation of narrative samples that can be compared across populations (Sealey and Gilmore, 2008), we review studies that investigated structured narratives, similar to ours, elicited with the support of pictorial stimuli.

In a study that elicited narratives relying on the wordless picture book, "Frog where are you?", from a group of 33 English-speaking children and young adults with Down syndrome aged between 12 and 26 years (mean age: 18.76), Miles and Chapman (2002) reported that expression of plot line was commensurate with non-verbal mental age and syntax comprehension. According to the report, thematic content was conveyed at a level consistent with their syntax comprehension as opposed to expressive language. However, when compared to TD controls matched on a measure of expressive language such as mean length of utterance (MLU), participants with Down syndrome expressed significantly more plot line events, thematic content and episodic events relating to character misadventures. Overall, the authors conclude that the narratives of participants with Down syndrome in this study indicate levels of conceptual abilities that are in line with syntax comprehension and non-verbal mental age, but tend to exceed expressive language ability. It appears, then, that macrostructural elements of narrative skill may not be accurately predicted by measures of expressive language such as MLU alone.

Finestack et al. (2012) examined the macrostructural story-telling skills of 24 English-speaking children and young adults with Down syndrome, aged between 12 and 23 years (mean age 16;9), compared to TD controls and individuals with Fragile X. Narratives were elicited using the wordless picture book "Frog goes to dinner." Participants with Down syndrome outperformed the non-verbal mental age-matched TD group on all macrostructural story components produced (Character Development, Mental States, Referencing, Conflict/Resolution, Cohesion, and Conclusion), with the exception of the Introduction component. When confronted with an MLU-matched control group, however, participants with Down syndrome did not show an advantage on any macrostructural dimensions.

In their investigation of a sample of participants that overlaps with that of Finestack et al. (2012) above, Channell et al. (2015) evaluated narrative abilities in 23 adolescents with Down syndrome aged between 10 and 16 years (mean age: 12). Participants with Down syndrome were reported to produce fewer episodic story elements compared to TD children matched for non-verbal cognitive ability. However, such difference was removed once MLU was controlled for, leading the authors to advance that individuals with Down syndrome do possess the necessary conceptual knowledge to express event-based story elements at a level consistent with non-verbal reasoning ability, but are limited in their expression of such elements by under-developed syntactic skills.

Zanchi et al. (2021) used an 18-picture storybook created to elicit the narratives of 13 Italian-speaking children and adolescents with Down syndrome, aged between 10 and 16 years (mean age: 12). No differences were observed on the

macrostructural elements in the stories of participants with Down syndrome and two groups of control participants, one matched on non-verbal MA (aged 5;2) and the other on MLU (5;5): all participants produced stories with comparable narrative structure and amount of event-based information.

The above studies typically control for MLU, or use MLU to match individuals with Down syndrome to control participants who are typically developing. However, while MLU is a useful measure of grammatical development in young TD children, its validity has been questioned in comparisons of grammatical mastery of children older than 4, both typically developing and in language-impaired populations (Scarborough et al., 1991). It is well-known that older children and young adults with Down syndrome will have richer vocabularies, producing more words in their utterances, but may still lack inflectional morphemes marking grammatical contrasts such as tense, which TD children acquire by about the age 4 (control participants in the studies above are aged 4 or 5). Studies relying on matching participants on vocabulary measures may thus provide richer insights into the narrative abilities of individuals with Down syndrome. The only study that focused solely on adults with Down syndrome and used vocabulary as a matching measure, Martzoukou et al. (2020), examined the story retelling abilities of 20 Greek-speaking adults with Down syndrome aged between 19 and 46 years (mean age: 28;2) by using two story retelling activities from the LITMUS-MAIN tool (Gagarina et al., 2012). The use of MAIN is of particular interest, as the task presents with several strengths. Firstly, MAIN offers a valuable tool for the controlled elicitation of narrative samples, while providing a detailed framework for the evaluation of macrostructural components of narrative ability. Furthermore, MAIN constitutes a particularly adept tool for the elicitation of expressive language in populations affected by developmental delays, as the picture-based retell activity in particular helps reduce cognitive load during narrative production (compared to those that use wordless picture books without providing a model story; Sealey and Gilmore, 2008). Secondly, since its development, the tool has been adapted to a variety of languages and cultural contexts, and has been adopted in a plethora of studies carried out all over the world to assess language abilities in typical and atypical populations (see https://main.leibniz-zas.de/en/worldwide-network/). As such, the task offers unique opportunities for cross-study comparisons and replications across a variety of populations. In their investigation of macrostructure, Martzoukou et al. (2020) report that individuals with Down syndrome performed worse on story structure relative to expressive vocabulary-matched TD controls, but no different to TD controls matched on non-verbal mental age. Comparison of participants' use of terms describing characters' internal states and emotions revealed that adults with Down syndrome used fewer such terms compared to both control groups. In addition, individuals with Down syndrome were found to perform similarly to non-verbal mental age matched controls and significantly worse than expressive vocabulary-matched controls on a series of comprehension questions tapping into the internal states and goals of characters in the story.

There are currently no studies reporting results from a remote collection of narratives from individuals with DS. Nonetheless, recent unpublished data suggest that this mode of elicitation of narratives is successful with young TD children (Sultana, 2022) and children with other developmental disorders such as autism (El-Raziq, 2022). Studies collecting natural language samples remotely provide reassurance that the quality of data obtained through parent-child interaction *via* video chat is of comparable quality to that obtained in the more traditional lab setting, for both TD children (Manning et al., 2020), and children with autism (Butler et al., 2022). Importantly, no differences in the crucial characteristics of TD children's samples such as speech intelligibility, lexical variety, grammatical errors or MLU were reported in samples obtained in person vs. video chat in Manning et al. (2020). Though such data are not yet available for individuals with DS, Kelleher et al. (2020) suggest that it is feasible to expect high quality data from unstructured parent–child interaction for infants with DS.

The current study

The present study examines the narrative language abilities of adolescents and young adults with Down syndrome by comparing them to a sample of TD controls matched on age of vocabulary comprehension. This represents part of a broader research project seeking to characterize the language skills of adults with Down syndrome and examine changes in language skills occurring over the course of adult life, particularly in relation to cognition and chronological age. With the current paper, we aim to establish a baseline for the general level of narrative language functioning achieved in early adulthood, by reporting on the abilities of a sample of young adults assessed before any suspected decline may have taken place. In the age range investigated here (15–35 years), cognition has not been affected by decline and language development is deemed to be approaching adult-like levels of syntactic organization and pragmatic proficiency. We expect this attainment to be reflected in our sample's story re-telling abilities. To elicit structured and comparable speech samples, we presented English-speaking participants with a story retelling activity from the Multilingual Assessment Instrument for Narratives (MAIN; Gagarina et al., 2019). In our study, the task was administered remotely during a video call with the participant and a caregiver. As such, in addition to being one of a handful of studies examining narrative language in adults with Down syndrome, the current study also represents the first attempt, to the best of our knowledge, at assessing narrative language skills remotely in a sample of people with Down syndrome. In this report we focus

on macrostructural aspects of narrative ability by examining performance on three dimensions of the MAIN retell task: Story Structure, production of Internal State Terms (IST), and Story Comprehension. We also analyse the narrative productions in terms of narrative length (total number of words produced) and lexical diversity (number of different words produced). The performance of our sample is compared to that of a control group of younger typically developing children matched on age of vocabulary comprehension. We asked the following questions about the abilities of our participant sample:

1. Will adolescents and young adults with Down syndrome produce narratives of length and lexical diversity comparable to younger vocabulary-matched TD controls?

2. Will adolescents and young adults with Down syndrome produce narratives with story structure, internal state terms and comprehension scores comparable to those of a younger TD control group matched on vocabulary comprehension?

3. Will the online approach be successful in eliciting narratives remotely and assessing verbal and non-verbal skills of individuals with DS?

In line with the previously reviewed literature on narrative skills in children and adolescents with Down syndrome, our participants should perform comparably well to matched TD controls on macrostructural properties of narrative retell. However, in line with the (sparse) literature on the narrative abilities of adults with this condition, it is also possible that our participants may perform less well-compared to verbal-MA matched controls, as reported in Martzoukou et al. (2020). Concerning the success of online elicitation with individuals with Down syndrome, while there is currently no literature on this topic, we expect that online assessment will be feasible based on recent literature with other populations with developmental disorders, such as autism.

Methods

Participants

Thirteen English-speaking participants with Down syndrome aged between 15;3 and 32;11 (M = 21.26) and 12 TD controls aged between 4;4 and 10;6 (M = 6.47 years) took part in the study. The chronological ages of TD children were matched to the verbal MAs of the participants with Down syndrome, as derived from the British Picture Vocabulary Scales-3 (BPVS-3) (see Ring and Clahsen, 2005, and Martzoukou et al., 2020, for similar matching methods).

Supplementary Table 1 shows demographic details of the two groups and descriptive background measures for the participants with Down syndrome: vocabulary comprehension (BPVS-3), grammar comprehension (TROG-2), non-verbal reasoning (KBIT-2) (see "Background measures" for more details).

All the participants with Down syndrome had a diagnosis of Trisomy 21. Two participants also had a diagnosis of ASD, while another had a diagnosis of obsessive-compulsive disorder (OCD) and bipolar disorder. None of the TD controls, all in primary school education, were reported to have any additional diagnosis or to be suspected of having any developmental delays.

As assessed by initial screening questions, six participants with Down syndrome had been diagnosed with hearing loss. Of these, four reported habitually using hearing aids. None reported other chronic illnesses or physical handicaps, and all reported speaking English as their main language. Two participants were reported to speak a second language non-fluently (Polish and Italian, respectively), while another three families reported speaking another language in the home (Konkani, Gujrati, French).

Four participants with Down syndrome were out of education at the time of testing, three attended secondary school and six were enrolled in further education. Participants with Down syndrome were reported to receive differing levels of support and attending various kinds of activities during the week. Some parents mentioned that their child received occupational and speech and language therapy (SLT) over the course of their lives, albeit irregularly, and no participant reported receiving regular ongoing SLT support. The lack of regular support at least for the past 2 years could be attributed partly to Covid-19, since many SLT services in the UK were significantly reduced during this time.

Participants with Down syndrome were recruited through online means. These included posts on social media platforms such as Facebook, and online adverts for the study shared by organizations such as the Down Syndrome Association (DSA). Though we did not collect specific information about where the participants heard about the study, Facebook posts seemed to be particularly fruitful when shared on groups dedicated to publicizing research opportunities for the Down syndrome community. A few of the participants were reached with the help of local charity organizations in London. Typical controls were recruited amongst contacts of the research team. Consent was provided by both participants and caregivers taking part. Participants with Down syndrome were provided with easy-read versions of the information and consent forms and TD controls completed child-appropriate versions of the form designed for different age groups. Most participants completed and returned the forms in digital version over email, while a small number requested a paper copy which was returned by post. Participant and caregiver were offered £10 vouchers each upon completion of the study.

The study was reviewed and approved by the ethical board of the Division of Psychology and Language Sciences at University College London (UCL).

Materials and procedure

The study involved a wide range of experimental measures and informant questionnaires which are not reported here. Below we provide the details and the administration procedure for both the LITMUS-MAIN narrative retell task and the three background measures that provide information about the general language and cognitive functioning of the participants in our sample.

LITMUS-MAIN narrative task

Narrative retell samples were elicited using the Cat story retelling activity from the revised version of the Multilingual Assessment Instrument for Narratives (MAIN; Gagarina et al., 2019), which was developed as part of the LITMUS test battery. In the current study, the task was adapted for online administration by following two previously developed adaptations: the first developed by Kapalková et al. (2021), for use with children, and another adaptation developed by Karl (2019) which was used with adults.

Our adaptation was presented on a PowerPoint presentation consisting of 28 slides, which we developed specifically to closely follow the administration guidelines from the original and revised face-to-face protocols (Gagarina et al., 2012, 2019). This task involves a scripted story which is presented alongside six pictures showing key events of the story. The narration involves four characters: a cat, a butterfly, a ball and a boy, and aims to assess macrostructural components of storytelling, such as structural complexity and use of internal state terms, as well as story comprehension. The activity involves three main parts: listening, retelling and comprehension. During the listening part of the activity, the participant is shown three colored envelopes and they are asked to pick one to reveal the story hidden inside. After opening one of the envelopes, the child looks at the pictures of the story which are initially presented on the screen silently. After examining the pictures, the child listens to the story, which is delivered in parallel with the relevant pictures. In our adaptation, participants all listened to the same pre-recorded version of the Cat story, which was presented in three sections, each accompanied by the relevant pictures. The story was recorded in advance by a female native English speaker. This was done in order to minimize variability amongst instances of task presentation in our participant sample. After listening to the story, participants are asked to retell the story. In our adaptation, the experimenter instructed participants by saying: "Now I want you to tell the story. Look at the pictures, and tell the best story you can. Imagine that you are telling the story to your favorite teacher who cannot see the pictures." These instructions were chosen to mimic the face-to-face protocol in which the experimenter doesn't share the story context with the participant (i.e., the experimenter cannot see the pictures). The adaptation intended to maintain this aspect of the task, as the availability of shared information can affect referential elements of children's retell (Gagarina et al., 2012, 2019). Consequently, we chose to address the retell toward a "favorite teacher," as we expected this to provide a friendly, but neutral figure, which also projects some elements of authority. For some of the adults with Down syndrome who were out of education this instruction was not applicable. In these instances, we asked them to "Imagine you are telling the story to a friend who cannot see the pictures." After hearing the instructions, the participant would begin telling their story as the experimenter moved through the pictures on the screen. As it was difficult at times to judge when the participant had completed the retelling of a portion of the story, the experimenter would check before moving to the next set of pictures by asking "Anything else?" The final part of the task assessed comprehension. In this portion, participants were shown the six pictures on one slide and asked questions about the story. For each question, the relevant pictures were highlighted by a red border, to focus the participants' attention to portions of the story relevant to the question. The activity was recorded for later transcription and scoring.

The adaptation used in the current study borrowed elements from the adaptations mentioned above, though it also included some additions. In particular, a few blank slides were added at intermediate points between the listening, retelling and comprehension portions of the task. This was done to ensure the participant was listening to the experimenter giving the instruction, rather than being distracted by the pictures on the screen. In particular, for the retelling portion, this addition ensured that the participant listened to the entire instruction before beginning to tell their story. A further addition included another blank slide placed at the beginning of the presentation which only contained an audio file playing a snippet of the recorded story (i.e., the speaker saying "1 day"). This was included to check that the sound from the presentation was audible to the participant through screen sharing before beginning the task. The slide was added after a few instances during task administration in which some participants were unable to hear the sound coming from the presentation once the first part of the recorded story was played out. The issue was most often due to the experimenter forgetting to tick the "Share sound" option when starting to screen share, while in a few cases it was due to an error of the PowerPoint presentation (e.g., presentation freezing). Slide numbers were also added to the presentation in order to facilitate administration for the experimenters.

Background measures

BPVS-3

Vocabulary comprehension was assessed using the British Picture Vocabulary Test, Third Edition (BPVS-3, Dunn and Dunn, 2009). The task was administered according to manual instructions and pictures from the stimulus book were shown

on screen using a visualiser camera. The video from the camera was screenshared with the participant by using the camera's visualiser application, as this allowed both the visualiser and the experimenter's camera to be active at the same time. During the teaching portion of the test (i.e., on Training plate A), participants' attention was drawn to the labels of the four pictures by saying: "Look, here are four pictures. There's picture One (pointing to the picture), picture Two (pointing to the picture), picture Three (pointing to the picture) and picture Four (pointing to the picture)." Whenever possible, participants were encouraged to verbalize their responses. The experimenter asked them to "Tell me which picture goes with the thing I have said." If participants tried to point or verbalized a label for the picture (e.g., "it's this one"), the experimenter would say "Could you tell me the number of the picture?" In some cases, the participants preferred to point rather than to verbalize their responses. In these instances, the experimenter asked the parent to assist and relay the number of the picture chosen by the participant.

KBIT-2 matrices

Non-verbal reasoning was assessed using the Matrices subtest from the Kaufman Brief Intelligence Test, Second Edition (KBIT-2, Kaufman and Kaufman, 2004). The assessment was administered as per manual instructions and pictures from the stimulus book were shown on screen by using a visualiser camera, in the same way as for the BPVS-3. The experimenter would administer the items by pointing to the relevant parts of the stimulus page as instructed by the manual. Again, participants were encouraged whenever possible to verbalize their responses by saying the letter associated with the chosen picture. If necessary, the experimenter would draw participants' attention to the labels of the response options by pointing out the letters associated with each picture during the teaching phase. If participants tried to point or verbalized a label for the picture (e.g., "it's the truck"), the experimenter would say "Could you tell me the letter of the picture?" In cases where participants preferred to point rather than verbalizing their responses, the experimenter asked the parent to relay the letter of the option chosen by the participant.

TROG-2

Grammar comprehension was assessed using the Test for Reception of Grammar, Second Edition (TROG-2; Bishop, 2003). The test was administered according to manual instructions and pictures from the stimulus book were shown on screen by using a visualiser camera, in the same way as for the BPVS3. In a similar fashion as for the BPVS-3, experimenters would point out the labels of the pictures during the teaching phase and encourage participants to verbalize their responses. When this was not possible, the parent would be asked to relay the number of the picture selected by the participant.

General procedure

All assessments were administered over a video call. Participants were invited to join a video call with the experimenter and were often accompanied by a parent, who sat next to them throughout the assessment and aided when required.

Data collection was typically completed over three video calls. The first video call arranged entailed completing the informant questionnaires with the parent or caregiver. The parental video call was typically scheduled first to create rapport with the parent and give them and the participant an opportunity to become more familiar with the researcher and ask questions about the study.

During the second and third video calls, participants completed the assessments administered by the experimenter. Participants with typical development completed the experimental measures during these sessions, while participants with Down syndrome were also administered the full battery of background measures (BPVS-3, TROG-2 and KBIT-2). The order of completion for both parental questionnaires and participant assessments was counterbalanced across participants: every other participant completed assessments in the reverse order, and the parent completed the questionnaires in the reverse order as well. When assessments were administered in the reversed order, BPVS-3 was moved to the beginning of the second session. This was done to maintain a standardized assessment at the beginning of each session, as this task provided a straightforward activity to engage the participant at the start of the call, before completing the more demanding experimental tasks.

Assessments were administered by the experimenters using a laptop or personal computer (PC) and a video camera (either USB or built-in webcam), using commercially available videoconferencing software (i.e., Microsoft Teams, Zoom). Experimenters wore headphones with a microphone during administration of the assessments. Administration of the standardized assessments was carried out using an OKIOLABS OKIOCAM T Compact A3 Visualiser/Document Camera.

Participants joined the testing sessions and completed the assessments using the equipment that was available to them in their home environment, typically either a laptop or tablet. In some cases, participants wore headphones, though the majority completed the assessments without them. This was especially the case for participants with Down syndrome: all but one of the participants preferred to not use headphones (these were sometimes not available, or the participant refused), though they always were encouraged to wear their hearing aids when available.

Scoring and data analysis

The speech samples collected during the MAIN retell activity were each transcribed by two separate independent transcribers.

Transcripts generated for each of the participants were compared to one another and conflicts resolved in accordance between the two original transcribers where possible, or by the first transcriber. Inter-transcriber reliability was checked for 50% of transcripts revealing an inter-rater agreement rate of 94–95%.

Coding and scoring of the narrative data were carried out by the first author. Scoring followed official guidelines from the MAIN assessment tool. The task is divided into three episodes, each involving two characters of the story. Each episode is structured to include five components: (1) an internal state generating the sequence of events (e.g., the cat saw the butterfly), followed by a (2) goal (e.g., the cat wanted to get the butterfly), followed by an (3) attempt (e.g., the cat jumped) and an (4) outcome (e.g., the cat fell in the bush). The sequence ends with an (5) internal state as a reaction to the outcome of the episode (e.g., the cat was hurt). Participants are awarded one point on story structure for each of the possible structural elements produced during their retell (up to 5 points for each episode). Two additional points are awarded for specifying the initial setting in terms of place and time (e.g., 1 day, at the lake). Following this scoring system, a score was generated for the number of structural elements present in the participants narratives. A score for use of internal state terms (ISTs) was also calculated by counting the total number of IST tokens produced by the participant during retell. Each use of words expressing perceptual states (e.g., look, see, hear), physiological states (e.g., hungry, tired, hurt), emotional states (e.g., sad, happy, scared), consciousness terms (e.g., awake, asleep), mental verbs (e.g., want, think, know), and linguistic verbs (e.g., say, shout, ask) was awarded 1 point. Finally, a comprehension score was obtained based on the participants' answers to a series of comprehension questions asked after retell, where each appropriate answer was awarded 1 point (for a maximum of 10 points). The comprehension questions are designed to tap into the participant's knowledge of characters' intentions and internal states.

As measures of narrative length and lexical diversity, respectively, the total number of words (TNW) produced by each participant during retell and the number of different words (NDW) used were also calculated. Total number of words included a count of all words produced by the participant during retell, with contractions (e.g., don't) being counted as two separate words. Only material relevant to the story was included in this count. Number of different words was calculated by counting each instance of different words occurring in retell, with same-stem words (e.g., run and ran, or want and wanted) being counted only in the first instance.

Statistical analyses were carried out in RStudio (RStudio Team, 2020, version 2022.02.3+492) using R (R Core Team, 2021, version 4.1.2). Parametric independent samples Student's *t*-tests were used to investigate mean differences between the groups on measures of story structure, comprehension, number of IST tokens, total words and number of different words. All assumptions of the Student's *t*-test were met. Besides *t*-statistics and *p*-values, we report effect sizes for each pair of comparisons as calculated using Cohen's *d* and interpreted according to the following commonly adopted guidelines (Cohen, 1988): small effect ($d = 0.2$); medium effect ($d = 0.5$); large effect ($d = 0.8$).

Two participants with Down syndrome (aged 27;2 and 30;4) were excluded from the analyses as they were not able to complete the MAIN retelling task, resulting in sample sizes of 11 participants with Down syndrome and 12 TD controls. An independent samples Student's *t*-test confirmed that there was no significant difference between the mean chronological age of TD participants and the mean vocabulary age equivalent for the participants with Down syndrome [$t(23) = -0.08$, $p = 0.937$].

Results

Supplementary Table 2 reports means (standard deviations) and ranges for Macrostructural (Story Structure and Internal State Terms) and Comprehension measures derived from the MAIN retell activity scoring, as well as means (and standard deviations) for narrative length (TNW) and lexis (NDW) produced by the two groups.

Statistical analyses revealed no significant difference between the groups on narrative length, as measured by total number of words produced during retell [$t(21) = -0.877$, $p = 0.39$, $d = 0.366$], however a significant difference in lexis emerged when comparing the number of different words produced by the groups [$t(21) = -2.30$, $p < 0.05$, $d = -0.961$]. Typically developing children produced a greater quantity of different words throughout their narratives relative to the participants with Down syndrome, despite overall length showing no difference across the groups.

Comparisons of macrostructural performance revealed a significant difference between the groups on the Story Structure score [$t(21) = -2.68$, $p < 0.05$, $d = -1.12$], with TD controls achieving higher scores compared to the participants with Down syndrome This indicates that TD children tended to include more elements of story grammar in their retells, such as initiating events, reactions, characters' intentions, actions and consequences. No difference, however, was found in the number of internal state term (IST) tokens produced by the two groups [$t(21) = -1.55$, $p = 0.135$, $d = -0.649$], suggesting similar rates of acknowledgment of characters' emotional, physiological or mental states across the groups.

Finally, a significant difference emerged when comparing Comprehension scores [$t(21) = -3.03$, $p < 0.01$, $d = -1.26$], indicating that participants with Down syndrome were outperformed by the TD group on our measure of story comprehension. This indicates lower overall accuracy of responses to questions in our study for the group with DS relative to our TD controls.

Discussion

The current paper presents the methodological approach and findings of a pilot study of narrative abilities in adolescents and young adults with Down syndrome. The study employed a picture-based retelling activity from the MAIN narrative task developed by Gagarina et al. (2019), as well as a range of background measures of language and cognitive ability, all presented online due to the COVID-19 epidemiological restrictions in place at the time of recruitment. The analyses included in this report sought to investigate aspects of macrostructural abilities, comprehension, narrative length and lexical diversity of story retell samples produced by a group of participants with Down syndrome aged between 15 and 33 years and a group of vocabulary age-matched TD controls. Due to the small sample size, the results should be interpreted with caution. However, considering the dearth of studies using online methodology to investigate language skills in this vulnerable population, we believe that our insights will provide valuable guidance for future research, particularly in relation to the assessment of language skills in the population with Down syndrome.

Our analyses revealed differences between the participants with Down syndrome and the vocabulary-matched controls in terms of both narrative structure and narrative comprehension. Thus, in answer to our second research question, our results clearly indicate that the adolescents and adults with Down syndrome, aged between 15;3 and 32;11, were outperformed by their much younger TD counterparts, aged 4;4 to 10;6. With regards to story structure, our control participants were able to produce more story components, i.e., Setting, Goals, Attempts, Outcomes, initiating events and reactions, in their retells compared to the adolescents and young adults with Down syndrome. This suggests that individuals with Down syndrome exhibit more difficulty with structural aspects of story-telling when vocabulary ability is controlled for. Such a difference may be attributed to overall expressive language deficits in the population with Down syndrome, widely reported in the literature. However, in addition to the recognized syntactic deficits (also confirmed by our participants' poor performance on the standardized measure of grammar comprehension, discussed in more detail below), it is possible that our participants were less skilled in producing relevant story components due to an additional presence of pragmatic deficits. Taking into account the perspective of the audience and the common ground shared between the speaker and listener is crucial in telling a successful narrative. Pragmatic difficulties have been observed in children with Down syndrome (Smith et al., 2017), despite early reports of relatively spared pragmatics (Roberts et al., 2007). A more detailed investigation of our participants' pragmatic abilities, independently of the narrative, may shed light on the nature of this finding.

The same issue is pertinent with regards to our second finding, our participants' poorer performance on story comprehension. Adolescents and young adults with Down syndrome provided fewer accurate answers than TD matches when asked a series of comprehension questions tapping into character's internal states and goals. The finding that participants with Down syndrome performed below vocabulary age expectations on the comprehension portion of the task fits with the widely reported weaknesses in sentence comprehension in this population. While the current paper does not focus on microstructural components of the narratives produced, our participants' poor comprehension score is in line with the literature highlighting sentence comprehension as a particular weakness in Down syndrome. As observed in their poor overall scores on the standardized measure of grammar comprehension administered in the current study, TROG-2, our participants showed significant difficulties interpreting a range of syntactic structures incorporated in this assessment. This is in line with existing literature showing floor scores on TROG-2 across different ages of individuals with DS (e.g., Frizelle et al., 2019), as well as studies focusing on specific complex syntactic structures that include passives (Ring and Clahsen, 2005; Perovic and Wexler, 2019) or relative clauses (Joffe and Varlokosta, 2007; Frizelle et al., 2019). However, it is not clear how much of the comprehension difficulty shown by our participants can be attributed to their grammatical difficulties, compared to possible difficulties in understanding mental states of the characters involved in the story, as intimated earlier.

Our analysis did not reveal differences between the groups on the number of internal state term (IST) tokens—words used to express the mental and emotional states of characters in the story—present in each group's narratives. This finding indicates that participants with Down syndrome produced words expressing internal states at a level comparable to TD controls matched on receptive vocabulary, suggesting that possible syntactic and/or ToM difficulties associated with the expression of mental states did not affect their production beyond vocabulary-based expectations. However, a larger sample of participants is needed to help us establish these facts: despite the absence of statistically significant difference between the groups, participants with Down syndrome showed lower performance on this measure. Nonetheless, a more granular analysis of the types of IST tokens and associated syntactic constructions produced by speakers in our groups may provide insights into their relative abilities to interpret and express the goals and emotions of others.

Interestingly, we found no evidence of a difference in narrative length between the two groups, as calculated by the total number of words produced by participants. This suggests that the difficulty exhibited by participants with Down syndrome in the production of structural components cannot be explained in terms of the raw length of their narratives alone. In other words, despite producing similar amounts of

words, participants with DS still mentioned fewer elements of the story structure during retell. While narrative length was similar between the groups, the number of different words used differed significantly: individuals with Down syndrome were outperformed in this respect, indicating that TD participants exhibited greater levels of lexical diversity compared to participants with Down syndrome. This may suggest that participants with DS tended to focus on selected aspects of the story to the exclusion of others, possibly reformulating or repeating information more often throughout their narratives.

Our results are not in line with those of studies that included younger participants with Down syndrome reviewed earlier. Recall that children and adolescents with Down syndrome (10−16 years, mean age: 12) in Zanchi et al. (2021) produced story structure and event-based information at a level commensurate to their expected non-verbal ability and MLU. Previously, Miles and Chapman (2002) have suggested that adolescents and young adults with Down syndrome (12–26 years, mean age: 18.76) produce narratives with structural elements, episodic events and thematic content that are in line with syntax comprehension and non-verbal cognitive levels, but which surpass what might be expected on the basis of MLU. A possibility, such as the one advanced by Channell et al. (2015), is that conceptual story telling abilities are relatively spared in Down syndrome, and at the level of non-verbal ability, but expression is limited by underdeveloped syntactic abilities. This is supported by Finestack et al. (2012) who also report an advantage on macrostructural story components when controlling for non-verbal mental age, but not when controlling for MLU. As noted earlier however, MLU may not be an appropriate measure for capturing expressive syntax abilities in adults with Down syndrome, as the informativeness of MLU as a measure of syntactic complexity significantly declines with age, and after an MLU of 4.0 (CA: 3 years) has been reached (Scarborough et al., 1991). Martzoukou et al. (2020) had speculated on the effect of world knowledge, associated with age, on the structural organization of narratives in Down syndrome. They hypothesized that the greater world knowledge of adults with Down syndrome would translate into better narrative structure compared to that of younger TD controls matched for expressive vocabulary and non-verbal mental age. However, this hypothesis did not find support in their analysis and does not seem to be supported by the analyses presented in our study, at least in terms of receptive vocabulary. Overall, our findings are partly in line with results reported in Martzoukou et al. (2020), the only other study employing the same instrument to assess narrative skills of adults with Down syndrome, though Greek rather than English-speaking, and administered in person rather than online. The results of their study also revealed poorer performance on both story structure and story comprehension from adults with Down syndrome (19–46 years, mean age 28;2) when compared to expressive vocabulary-matched TD controls. In terms of use of IST tokens, our study found no differences in the frequencies of internal state terms use between the two groups, while Martzoukou et al. report significantly fewer ISTs in the productions of their participants with Down syndrome compared to both of their TD control groups, one matched on expressive vocabulary and the other on non-verbal ability. Martzoukou et al. interpret the lower frequencies of mental state terms in the narratives of participants with Down syndrome as a result of their poor syntactic ability: since verbs used to express internal state terms almost always require complex syntactic constructions such as complement clauses (de Villiers and de Villiers, 2009), it may be this syntactic complexity that precludes use of mental state terms in individuals with Down syndrome. However, this difference in IST use might alternatively be explained by the relatively older age of the sample recruited by Martzoukou et al. It is possible that, given the ages reported in their study, some of the participants included in their sample might have already started to experience symptoms of cognitive decline which reflected on their language performance. Crucially in our sample, we include participants below the age of 35, a stage of life at which we expect language development to be approaching adult form. We would expect individuals with Down syndrome between the ages of 15 and 35 to show degrees of variability in their relative skills, across both language and cognition, as performance in this population is often reported to be highly heterogeneous (Roberts et al., 2007). However, we wouldn't expect neuropsychological symptoms of Alzheimer's disease to influence performance in this age range, as the impact of dementia-related decline becomes manifest around the fourth decade of life (Ballard et al., 2016). Based on this, we would expect the abilities of our sample to be reflective of adult macrostructural narrative abilities, unaffected by decline.

With regards to our third research question, in this study we were able to show that diverse types of data can be successfully obtained *via* online remote administration from individuals with Down syndrome. Our results suggest that measures of general language and cognitive abilities can be used remotely with the population with DS. Only two participants with Down syndrome (aged 27;2 and 30;4) were excluded from the analyses reported as they were not able to complete the MAIN retelling task, however, they completed the background measures. These participants' inability to complete the retell task was primarily due to limited expressive language abilities and use of prompting from the caregiver during task administration.

All participants completed the grammar and vocabulary comprehension tasks, in addition to the task assessing non-verbal reasoning. Here we discuss how our participants scored on these very same measures as administered online, compared to those reported in previous literature, but administered face-to-face. While direct comparisons cannot be made, due to different age ranges of participants involved, the mean scores on BPVS-3 and TROG-2 for our sample of participants seem in line with those reported for in person assessments in the literature, suggesting that that online assessment of vocabulary

and grammar may be viable in the population with Down syndrome. Our participants' scores on TROG-2 are similar to those reported in the previously reviewed Finestack et al. (2012) for adolescents and young adults with Down syndrome aged between 12 and 26 mean age 18.76. The mean TROG-2 raw score reported in their study, 2.63 (SD = 1.58), range 0–6, is comparable to that seen in our participants, mean 3.38, SD (2.53), range: 0–7. With regards to BPVS-3, adults with DS from a LonDownS Consortium study (Startin et al., 2019b), aged between 19 and 59 (mean age 36.47 years), obtained a mean raw score of 95.94 (SD: 31.99) (range: 38–158), which is comparable to our participants' mean score of 89.23 (26.03) range: 53–119. Similar levels of non-verbal ability have also been previously observed on KBIT-2: in another LonDownS Consortium study, Startin et al. (2016) report mean non-verbal raw score of 14.98 (6.90), range 0–32, for a large sample of young healthy adults with DS aged 16–35 years (mean age 25.24 years), which is in line with our sample's mean score of 15.31 (5.06), range: 2–21. In addition, the finding that narrative length was comparable across our participant samples when matched on age of vocabulary comprehension suggests that the online elicitation approach was similarly effective in eliciting narrative discourse from both participants with DS and TD controls remotely. Further analyses of the initial data presented in the current paper will explore the nature of this finding in more details, as our results suggest that despite producing a comparable number of words, participants with DS included fewer elements of story structure and used a more limited range of different words in their narratives. We provide a more detailed discussion of our general experience with the remote collection of data in the section below, to allow future researchers to make informed choices when considering remote methods of assessment.

Methodological considerations on remote administration of experimental materials

Online methods of data collection in the population with Down syndrome present both advantages and disadvantages which must be taken into consideration when designing remote research approaches. Online methods minimize the need for travel and significantly reduce contingencies associated with the costs and time-investments of travel, for both researchers and participants. Furthermore, they allow for a relatively comfortable mode of testing, thanks to the commercial availability of numerous videoconferencing tools which have seen a significant uptake in usage in recent times, especially due to the social-isolation restrictions put in place all over the world. Participants can then be tested at home, from a familiar environment, while reducing possible health risks associated with face-to-face contact.

In terms of participant engagement, the pilot has yielded promising results so far. Our experiences do not confirm worries relating to participants' ability to engage in online activities. The families involved in the study were always able to connect to the videocalls using their electronic devices. In some cases, it was the child or adult participant who provided help to the parents, who were sometimes less familiar with online videoconferencing tools. In other cases, participants did require help from a parent to join the calls (this was virtually always the case for the TD children, but also for some of our participants with Down syndrome), but they were nonetheless able to engage with the experimenter and participate in the online tasks. For two participants with Down syndrome, the narrative task presented particular challenges, likely associated with more pronounced impairments in expressive skills. We speculate that the task itself may have been too demanding for these participants, however, it cannot be excluded that a face-to-face setting may have facilitated their performance. Future research would benefit the field by examining performance differences in people with intellectual disability between face-to-face and remote online task administration. As for recruitment, we again cannot exclude that the online nature of the study might have dissuaded some families from participating, however, we can report that a number of families expressed relief at the notion of being able to participate from home. Such relief might be in part associated with the relative ease of remote participation on the side of families and seems reasonable given the broader global context in which data collection has been taking place. In this respect, remote online assessment represents a promising new approach to data collection, especially when working with extremely vulnerable populations such as people with Down syndrome. Furthermore, an added benefit of the remote approach to assessment was that of allowing us to reach and involve participants and families based all over the United Kingdom, while significantly reducing traveling costs incurred by researchers.

Alongside the promising outcomes for both recruitment and data collection, we must also consider some of the drawbacks of remote online assessment. One of the main issues involve the representativeness of our sample, due to possible disparities in accessing the necessary technology to take part in online studies. While the global pandemic forced schoolchildren and students all over the world to participate in online schooling during official lockdowns in 2020 and 2021, forcing them and their caregivers to invest into technology, this was not possible for many families who faced economic hardship (UNICEF, 2021).

With regard to the experimental set up itself, the central issues revolve around a loss of experimental control over the surrounding environment, the equipment used, and a lack of direct influence over the engagement of the participant. In our pilot study, we observed significant variation in terms of the equipment available to the participants, as they completed the sessions from their home environment. Participants typically

completed assessments using a laptop or tablet, in most cases without wearing headphones, as these were uncomfortable to them or not available to the families. The majority of participants, across both groups, completed the assessments with a parent or caregiver present who provided assistance when necessary, without interfering with the assessments. Variations in the type of equipment available to participants is an important consideration in the design of online elicitation studies, as it may play a role in how the participants are able to engage with the tasks: for example, features such as screen size and resolution of the electronic device used could affect the visibility of the materials presented.

Most participants were able to complete the assessments following instructions given by the experimenter and did not require further assistance from the caregiver. In some cases, the caregiver helped by redirecting the participant's attention toward the tasks when distracted by environmental stimuli (e.g., noise, other family members in the house). In other cases, specifically with two participants with Down syndrome, the help of the parent was actively required in administering the background assessments, as the participants chose to point to pictures rather than verbalizing their responses. Given that the assistance, or at the very least the presence, of a caregiver is likely necessary during task administration, we recommend that future studies take measures to instruct the parent on how to behave and assist during the assessments, in order to avoid interference that can invalidate the quality of the data collected.

Another potential issue of remote online assessments, especially relevant when eliciting language samples, relates to the audio quality and intelligibility of the recordings collected. In this study, we collected language samples from the MAIN retell activity (Gagarina et al., 2019), which were subsequently transcribed and scored. For most participants, the reduced quality of language samples collected without using headphones did not have a significant impact on the experimenters' ability to transcribe the samples. However, this was not always the case for participants with Down syndrome, for whom in a few cases, low audio quality coupled with intelligibility issues significantly affected the ease of transcription. Our inter-transcriber reliability rates were excellent, however, providing families with equipment (particularly headphones and a microphone) could contribute to minimizing audio quality issues and may improve the intelligibility of the speech samples collected. One important drawback of using headphones, however, is that the parent or caregiver assisting with the session would not have auditory access to the instructions given by the experimenter. A further aspect that merits consideration relates to hearing difficulties, which appear to be common in the population with DS (Shott, 2000). In our study, six participants reported some degree of hearing loss which could affect their task performance. Of these, two were excluded from analysis as they failed to complete the retelling activity, while the remaining four wore hearing aids during task administration. While the use of hearing aids may help minimize the impact of hearing difficulties, remote language elicitation designs should seek to account for this factor: though this wasn't included in the present design, language research would benefit from the adaption of a hearing screening procedure for remote use.

Finally, we experienced minimal issues with regards to connection quality during videocalls, another area of uncertainty when administering assessments online. Participants overall seemed to have access to good enough internet connection to allow completion of the activities in the absence of signal degradation that could significantly affect task administration. Occasional issues were observed in terms of audio quality, with a few instances of audio glitches. Because the assessments required the participant being able to hear speech produced by the experimenter, we allowed experimenters to repeat items if the participant indicated that they did not hear the experimenter's prompt, though this was seldom the case for the tasks presented above (in particular, this was especially relevant for language comprehension tasks such as BPVS-3 and TROG-2).

Conclusion

The current research contributes to the growing body of literature documenting the language skills of adults with Down syndrome, a relatively understudied demographic, by focusing on macrostructural narrative language elicited by means of a novel methodological approach. In the first entirely remote study of narrative language abilities in adults with Down syndrome, we adapted the assessment tool for remote online use over videoconference. In addition to contributing valuable insights into the feasibility of remote online research designs with participants with intellectual disability, the study is also the first to assess narrative language in a group of adults with Down syndrome at an age range where language abilities are approaching adult performance, while unlikely to be affected by cognitive deterioration associated with Alzheimer's disease. We report a disadvantage of participants with Down syndrome relative to TD controls matched on age of vocabulary comprehension on global measures of story structure and story comprehension, as well as lexical diversity, though the groups did not differ on story length as measured by total number of words. In the current report, we discuss the implications of such findings in relation to previous literature assessing macrostructural narrative skills of children and adolescents with Down syndrome, as well as the more sparse evidence available on adults, and reflect on the outcomes of our remote online approach to language assessment. We conclude that remote online methodological approaches are viable tools of eliciting speech samples and assessing expressive language skills in adolescent and young adults who have Down syndrome.

Data availability statement

The datasets presented in this article are not readily available because this dataset includes data from vulnerable participants. Requests to access the datasets should be directed to Elisa Mattiauda, elisa.mattiauda.18@ucl.ac.uk.

Ethics statement

The studies involving human participants were reviewed and approved by UCL Research Ethics. Written informed consent to participate in this study was provided by the participants and their legal guardians. Child participants provided assent through a child-friendly form.

Author contributions

EM, AH, and AP designed the study. EM collected and analyzed the data and wrote the manuscript. AP contributed to the writing of the manuscript. AH edited the manuscript. All authors approved the final version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fcomm.2022.841543/full#supplementary-material

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