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REVIEW ARTICLE

Narrative Skills of Youth with Down Syndrome: a Comprehensive Literature Review

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Abstract Given the role that narratives play in organizing experience, as well as their contribution to social interaction and literacy development, children's narrative skills have received considerable research attention. This paper provides a comprehensive review and thematic analysis of 30 studies conducted over the last three decades on the narrative skills of children and adolescents with Down syndrome. Themes include the global structure of their narratives, narrative comprehension, and use of vocabulary and grammar. The review demonstrates that youth with Down syndrome have basic narrative skills, generally consistent with individuals of the same mental age or language ability, suggesting a delayed rather than an atypical pattern of development. The narrative profile emerging from the review indicates several ways narratives may be facilitated in children and adolescents with Down syndrome. These include a greater focus on personal narratives accompanied by the use of visual supports. The review revealed very few intervention studies, suggesting an important direction for future research.

Keywords Down syndrome · Narrative · Language · Storytelling · Story comprehension

Oral narratives have often been described as a primary and universal way that people organize and make sense of their experience (Polkinghorne 1988; Wells 1986). Indeed, narratives or 'stories' of past, future, or imagined events are ubiquitous in our daily lives; they are present in conversations, ceremonies, books, television, and play, ranging from early pretend to video games. Thus, the ability to understand and tell stories has an important role in social and communicative functioning. Moreover, given that stories are often read aloud by educators in early childhood settings and expected to

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be read independently by students later on, story knowledge can contribute to children's classroom participation and their academic performance.

For many children, narrative skills emerge at a young age, and are supported by exposure to narratives and adult scaffolding (Fivush et al. 2011). For children with developmental disabilities, narrative skills often develop slowly, due to difficulties with language and/or cognition. This appears to be the case for children with Down syndrome (DS), whose skills are weaker than typically developing peers of the same age (Bernicot et al. 2003; Lacroix et al. 2007; Reilly et al. 1990). However, knowing only that skills are below age expectations is insufficient to guide instruction. In order to assist children with DS to become competent narrators and to better understand stories in their everyday lives, more information is needed.

In the present review, we provide a portrait of narrative skills of individuals with DS in childhood and adolescence and present the implications of the findings for classroom instruction or specialist (e.g., special educator or speech-language therapist) interventions. To the best of our knowledge, this is the first review of its kind. The studies reviewed include comparisons of youth with DS to children matched on chronological age, as well as to individuals with other disabilities (e.g., autism) or to younger children, matched on mental age, language, or reading. We also review the rare studies which investigate narrative skills longitudinally and discuss response to intervention. Before presenting the results, we briefly discuss the significance of narratives in children's lives, describe DS, and present critical dimensions of narrative.

The Importance of Narrative in Children's Lives

Children's narrative skills merit attention for numerous reasons. First, personal narratives play an important role in children's emotional and social lives (Miller et al. 1990). Specifically, adult-scaffolded narratives contribute to children's memories about their own experiences, which, in turn, relates to children's early sense of identity (Fivush et al. 2011). Later, children use narrative to achieve interactional goals: for example, to create and maintain intimacy with peers (Engel 2005), to garner teacher attention and approval (Küntay and Senay 2003), and to organize and sustain pretend play (Ahn and Filipenko 2007). Additionally, narratives continue to play an important role in the interactions of older children, as they negotiate social organization and group belonging (Pesco and Crago 1996).

Narrative skills also relate to school achievement. They are included in the competencies expected of children at school (e.g., Ministry of Education, as cited in van Bysterveldt et al. 2012), particularly those pertaining to language arts activities. Oral narrative skills are also associated with reading comprehension (e.g., Griffin et al. 2004), a finding one can attribute to commonalities between oral and written narratives. Specifically, 'mature' oral narratives link events causally (Trabasso and Magliano 1996), involve information that needs to be inferred by listeners (van Kleeck 2008), and follow certain structural patterns (Schneider et al. 2005). Children may glean knowledge about these and other features of the narrative genre from exposure to oral narratives, which they can later evoke and extend to understand the texts they read. This point is relevant to children with DS, some of whom learn to read (Trenholm and Mirenda 2006). For non-readers, oral narrative skills will nonetheless impact their

abilities to participate in Language Arts activities. Listening to and telling stories can also help children acquire more language, and improved language, in turn, can have positive effects on children's behaviour (Buckley and Le Prèvost 2002).

Down Syndrome

DS is a developmental disorder resulting from an error in cell division (Finestack et al. 2012). Ninety-five percent of cases involve an extra copy of chromosome 21 (trisomy), while in the remainder, either the extra copy occurs in some cells (mosaicism) or parts of chromosome 21 attach to another chromosome (translocation) (Martin et al. 2009). DS is associated with particular physical features (e.g., distinct facial characteristics, low muscle tone); medical conditions including congenital heart disease; and intellectual disability, with IQs typically ranging from 30 to 70 (Chapman and Hesketh 2001).

DS is the most common genetic cause of speech and language disabilities (Rice et al. 2005). Speech problems in DS have been attributed to anatomical differences in the vocal tract and periodic hearing loss due to frequent otitis media (Rice et al. 2005). Dysfluencies in speech (i.e., frequent hesitations and repetitions) have also been observed, and attributed to difficulties in planning speech (Kover and Abbeduto 2010).

With respect to communication and language, infants with DS show delays in pre-linguistic communication and reciprocal eye contact, despite having high levels of social interest (Abbeduto et al. 2007). Nonetheless, some pragmatic strengths have been observed. Martin et al. (2009), for example, report that children with DS use a variety of communicative functions, stay on topic, and respond to requests for clarification. Receptive vocabulary has also been reported as a relative strength compared to various aspects of expressive language (Chapman et al. 2000; Cleave et al. 2012; Miles and Chapman 2002). Oral language skills have additionally been linked to reading. More specifically, receptive language predicted reading growth in a group with DS receiving reading intervention (Burgoyne et al. 2012).

Finally, individuals with DS have been found to display better visual-spatial memory than verbal memory (Campbell et al. 2013; Jarrold and Baddeley 2001). This relative strength has been capitalized upon in narrative research, both to elicit narratives (as discussed immediately below) and facilitate story comprehension (Lecas et al. 2011).

Dimensions of Narrative

Definition and Type

Narrative has been defined in various ways in the literature. Most agree that narratives involve an account of events connected in time. As elaborated below, however, theoretical models of narrative add other defining features. Narratives have also been distinguished as being either personal or fictional. In personal narratives, the narrator recounts actual events in which they were themselves involved. While personal

narratives arise spontaneously in conversation, they have been elicited in research contexts using verbal prompts (e.g., by telling a story then asking the participant "Did that ever happen to you?") or photographs of either everyday events or relating to the child's own experience. The latter strategy has been used in studies of individuals with DS (Chapman et al. 1998, 2000; Miles and Chapman 2002; van Bysterveldt et al. 2012).

Research participants, in the studies reviewed here and in narrative studies generally, are often asked to generate fictional stories while viewing a set of pictures (e.g., Miles and Chapman 2002) or moving images (e.g., Boudreau and Chapman 2000; Chapman et al. 1998, 2000). In other studies, participants listen to a story while viewing related stimuli (e.g., pictures, television), and are then asked to retell the story (e.g., Kay-Raining Bird et al. 2004), or to respond to comprehension questions (Kim et al. 2008).

Macrostructure

Narratives in DS have been analyzed largely in terms of their *macrostructure*: that is, in terms of elements included in the narrative and their organization. Two widely known models of macrostructure—story grammar and high point—are described below, followed by a discussion of theme, causal relationships, and inferencing, which also contribute to story structure and pertain to narrative skills in DS.

Story Grammar Story grammars define the elements that constitute a well-formed story or "episode" within a story. While various story grammars have been proposed since the late 1970s, they typically include information about characters, place, and time; initiating events (i.e., events that set others in motion); a problem or goal to be achieved by the protagonist(s); plans or attempts to resolve the problem or achieve the goal; consequences of those actions; and character responses, including internal states (emotions and mental states) (Schneider et al. 2005). According to story grammar models, these elements combine to create a schema (an internal representation of narrative structure) that, in turn, facilitates story recall, comprehension, and production (Berman 2009).

High Point and Evaluation High point stems from a model of narrative structure proposed by Labov and Waletzky (1967) which applies to narratives of personal experience. The elements in the model are broadly parallel to story grammar, in that they relay information about setting and events, but evaluation is an added and critical element. Evaluation involves the aspects of a story the narrator accentuates, including references to characters' internal states, use of dialogue, and stress and intonation; it signals the story's climax or "high point" and reveals which events are salient and presumably, meaningful, to the narrator (Reilly et al. 1990).

Theme While story grammar and high point focus on story plot, other research addresses story theme. According to Williams et al. (2002), understanding a story's theme involves 'going beyond the story' to make general value judgments (e.g., "Stealing is bad") or observations (e.g., "Some people steal"). In studies involving individuals with DS, theme has been defined more narrowly as simply the central problem of the story (Reilly et al. 1990).

Causal Relationships and Inferencing

In order to tell a comprehensible story, a narrator must either recount events in the order they occurred, and/or signal a reordering of the chronology (Labov and Waletzky 1967). In addition, narrators link events by making causal connections: events and internal states lead to or result from others (Trabasso and Magliano 1996). Causal connections may be explicitly stated, but often the listener or reader must infer them, drawing on world knowledge (van Kleeck 2008). Other kinds of inferences also contribute to understanding stories. These include "text-to-text" inferences that connect words in the text (e.g., a noun and a related pronoun) (van Kleeck 2008), as discussed further under cohesion in the next section.

Microstructure

Narratives have also been described and analyzed in terms of vocabulary, grammar, and cohesion, i.e., the ways narrators link sentences to one another or understand such links. These features have been referred to as the *microstructure* of a narrative (Finestack et al. 2012). These features are not unique to narrative; rather, they are linguistic skills observed in expression and comprehension generally. Given this, the emphasis in our review is on studies examining the production and understanding of the macrostructural features described above. However, microstructural features are described briefly in the results in order to give the reader a sense of the linguistic abilities of individuals with DS, as expressed in a narrative context. Below are some of the linguistic variables included in various studies.

Vocabulary In a narrative context, one could assess expressive or receptive vocabulary, or both. Expressive vocabulary measures in studies of DS often include *number of different words,* an indicator of vocabulary diversity.

Cohesion Cohesive devices connect sentences to one another. In studies of DS, cohesion measures have included (a) expressions that narrators use to refer to characters initially and throughout the narrative (e.g., using nouns initially and following them with pronouns) and (b) conjunctions that link parts of the narrative (e.g., "and then" to connect one sentence to another) (Boudreau and Chapman 2000).

Grammar Grammar entails both syntax and morphology. While syntax encompasses the rules for forming sentences, morphology involves the rules for forming words. Studies of DS have often employed the following widely used measures of grammar: *mean length of utterance* in morphemes (MLU) (e.g., the utterance 'I walk-ed' would have 3 morphemes) (Boudreau and Chapman 2000), and the use or understanding of various sentence structures (e.g., simple and complex clauses) (Fabbretti et al. 1997). Finally, narrative length has also been considered as an indicator of either expressive syntax or expression more generally. Length has been calculated in terms of total number of utterances (or other units, e.g., clauses) and in terms of volubility, such as the number of words per minute (Chapman et al. 1998, 2000).

Method

Search Strategy and Selection Criteria

To identify literature related to narrative skills in DS, we searched the following electronic databases: ERIC, PsycINFO, Medline, and LLBA. We limited the search to peer reviewed publications in English or French given our language expertise. We set no limits on publication date, in an effort to identify the earliest studies appearing in the databases. This search strategy resulted in studies from 1985 to 2013. We placed no limits on methodology, thus allowing for a range of study types. We included studies involving children and adolescents, and allowed studies with young adults only if these other two groups were included. We planned to exclude studies in which individuals with DS also had a comorbid developmental disability (such as autism or ADHD), or psychiatric disorders (however, none were identified). The search terms were ("Down syndrome" or "Down's syndrome" or "Down* syndrome" or trisom*) AND (narrat* OR story* OR stories).

The search yielded 83 unique references (duplicates were removed). The abstracts were screened against the following inclusion criteria: (a) entailed assessment of narrative skills of children or adolescents with DS and (b) included amongst the dependent variables one or more of the following: macrostructural elements (e.g., story grammar), theme, causal relations, inferencing, or microstructural features assessed in a narrative context versus some other discourse context (e.g., conversation). Thus, studies were excluded if they were either clearly irrelevant to the review's central concerns (e.g., narrative accounts of the experience of parenting a child with DS) or examined aspects of language or fluency using a narrative task, but without an express interest in narrative skills (e.g., Finestack and Abbeduto 2010).

Coding and Reliability

The second author screened all 83 abstracts. The first author then independently reviewed the same set of abstracts. Reliability for the inclusion decision (include or exclude) was 95.2 %; the four disagreements were resolved through discussion between coders and examination of the full texts. After applying the inclusionary and exclusionary criteria, 30 references remained: 18 focusing on narrative macrostructure and 12 on microstructure.

For macrostructure, the focus of our review, intercoder reliability was established. The first author coded half of these and the second author coded the other half in terms of groups; participant ages; sample size; outcome variables; and findings. To assess intercoder reliability, the two authors independently coded a third of the studies (selected at random). Reliability, calculated in percent agreement, was as follows: age and sample size 96.88 % (with disagreements stemming entirely from rounding or recording errors that were later corrected); groups 100 %; and outcome variables 96.67 %. The reliability for the report of findings was 81.25 %, with the vast majority of disagreements arising from differences between coders in how exhaustively the results had been reported. Disagreements were resolved through joint review and discussion of the studies. The authors also went on to jointly review the remainder of studies (i.e., those not in the reliability pool) until 100 % agreement was reached for the findings.

For microstructure, the first author recorded and verified the results; this was sufficient given that microstructure categories (e.g., vocabulary, MLU, syntax) are

reported in standard ways, while the terminology for macrostructural narrative features varies considerably across studies and thus required coding.

Results

General Description of the Studies

After applying the inclusionary and exclusionary criteria, 30 references remained. Most of the studies examined fictional narratives. Exceptions were Chapman et al. (1998, 2000), and Miles and Chapman (2002), who investigated both fictional and personal narratives, and van Bysterveldt et al. (2012), who examined only personal narratives. In nearly all of the studies (except Kay-Raining Bird et al. 2004), pictures or other visual prompts were used to elicit narratives.

Study designs often entailed comparisons of participants with DS to other groups, such as (a) typically-developing, younger, individuals matched on nonverbal "mental age" (hereafter, MA matches), language, or reading, and/or (b) individuals with other developmental disabilities, including Fragile-X syndrome (FXS), autism, and William Syndrome (WS). In contrast, three articles reported results uniquely for individuals with DS, examining the effects of time (Cleave et al. 2012) or contextual factors (Kim et al. 2008; van Bysterveldt et al. 2012) on narratives.

Macrostructure

Table 1 summarizes the groups of study, outcome variables, and significant findings for each study. The table presents only the outcome variables related to macrostructure, consistent with the focus of the review. Fifteen studies are included, but some appear more than once given that they examined multiple variables. Another three studies that involved only descriptive data (i.e., no between or within-group comparisons) are discussed in the text: Lecas et al., Schoenbrodt et al. and van Bysterveldt et al.). Also, as previously mentioned, the microstructure variables are covered uniquely in the text.

Story Grammar Bernicot et al. (2003) reported that children and adolescents with DS (expectedly) had poorer story grammar compared to chronologically aged (CA) matches and weaker skills compared to a group with WS. Three studies reported that children and adolescents with DS were similar to children matched by either mental (MA) or reading age in their recall and production of story grammar elements (Estigarribia et al. 2011; Hogan-Brown et al. 2013; Kay-Raining Bird et al. 2008). More specifically, Kay-Raining Bird et al. (2008) found that when children and adolescents with DS and a reading-matched group retold a story, they produced a comparable number of story episodes, regardless of modality (oral, word-processed, and handwritten). Hogan-Brown et al. (2013) studied children and adolescents with DS, younger language (vocabulary) matches, individuals with FXS (with and without autism) and autism alone. They examined the effect of a "macrostructure" variable, which included story grammar (specifically, number of main episodes), the main

Table 1 Narrative macrostructure of children and adolescents with DS

Authors	Groups (n)[a]	Age Range, Means (rounded up)	Outcome variable	Findings (comparisons involving DS groups)[b]
			STORY GRAMMAR	
Bernicot et al. 2003	1) DS (n=7) 2) WS (n=7) 3) TD (n=7)	7–19 years, M=13 7–19 years, M=13 7–19 years, M=13	Story structure (schema)	DS < TD-CA DS < WS
Estigarribia et al. 2011	1) DS (n=33) 2) FXS-O (n=29) 3) FXS+ASD (n=28) 4) MA matched to FXS (n=39)	6–16 years, M=11 6–16 years, M=12 6–16 years, M=12 3–8 years, M=5	Story structure Story structure subscales	DS = FXS-O DS = FXS+ASD DS = FXS+ASD
Finestack et al. 2012	1) DS (n=24) 2) FXS (n=12) 3) MA matched to DS on group MA, individual MA, plus subset matched to DS on MLU (n=21)	12–23 years, M=17 11–20 years, M=15 4–7 years, M=5	Story structure total Story structure subscales: introduction, character development, mental states, conflict/resolution, cohesion, conclusion	DS > MA-group match DS > MA-individual match DS = MLU DS = FXS DS > MA-group match introduction, trend on others DS > MA-individual match conflict/resolution, cohesion DS = MLU DS = FXS
Hogan-Brown et al. 2013	1) DS (n=17) 2) MA (n=16) 3) FXS (n=18) 4) FXS-ASD (n=23) 5) ASD (n=20)	7–15 years, M=11 3–6 years, M=5 7–15 years, M=10 6–15 years, M=10 4–13 years, M=9	Story structure	DS = MA DS = ASD
Kay-Raining Bird and Chapman 1994[c]	1) DS (n=47) 2) MA (n=47)	6–21 years: $M_{Time\ 1}$=12 2–6 years: $M_{Time\ 2}$=4	Sequencing recall Sequencing errors	DS < MA DS = MA

Table 1 (continued)

Authors	Groups (n)[a]	Age Range, Means (rounded up)	Outcome variable	Findings (comparisons involving DS groups)[b]
Kay-Raining Bird et al. 2004[c]	1) DS (n=23)	13–20 years, M=16	Proposition recall overall	DS < MA
	2) MA (n=24)	4–6 years, M=5[4]	Verbatim recall immediate	DS < MA
			Verbatim recall delayed	DS = MA
			Gist recall (see inferencing below)	DS < MA
			Extraneous information in story	DS > MA
Kay-Raining Bird et al. 2008[c]	1) DS (n=20)	9–20 years, M=14	Story structure (episodic structure)	DS = TD-RM
	2) TD-RM - reading match (n=17)	5–11 years, M=7		
Lacroix et al. 2007	1) DS (n=12)	7–19 years, M=12	Story structure (schema elaboration)	DS < MA
	2) WS (n=12)	7–19 years, M=12		DS < TD-CA
	3) TD-CA (n=12)	7–19 years, M=12		DS < WS
	4) MA-overall IQ (n=12)	4–10 years, M=7		
Loveland et al. 1990	1) DS (n=16)	5–27 years, M=13	Story structure (event recall);	DS = ASD
	2) ASD, matched to DS on MA-verbal (n=16)	5–27 years, M=14	Story character/object mentions; number of words in stories	
			Story comprehension (factual; internal states)	DS = ASD
			Story comprehension inappropriate responses	DS < ASD
Miles and Chapman 2002[c]	1) DS (n=33)	12–26 years, M=19	Story structure (total elements)	DS < MA
	2) MA (n=33)	2–9 years, M=5		DS > MLU
	3) MLU match (n=33)	2–9 years, M=4		DS = SYN-COMP
	4) SYN-COMP match (n=33)	2–8 years, M=5	Story structure (attempts to reach goal by boy character)	DS = MA
				DS = MLU
				DS = SYN-COMP
			Story structure: attempts to reach goal by dog character and story plot line	DS = MA
				DS > MLU
				DS = SYN-COMP

Table 1 (continued)

Authors	Groups (n)[a]	Age Range, Means (rounded up)	Outcome variable	Findings (comparisons involving DS groups)[b]
			EVALUATION	
Bernicot et al. 2003	1) DS (n=7) 2) WS (n=7) 3) TD (n=7)	7–19 years, M=13 7–19 years, M=13 7–19 years, M=13	Evaluative devices (attention-getters, emotional verbs, sound effects, dialogue)	DS = TD-CA DS < WS
Hogan-Brown et al. 2013	1) DS (n=17) 2) MA (n=16) 3) FXS (n=18) 4) FXS-ASD (n=23) 5) ASD (n=20)	7–15 years, M=11 3–6 years, M=5 7–15 years, M=10 6–15 years, M=10 4–13 years, M=9	Evaluative devices: use and diversity	DS = MA DS = FXS-AUT
Keller-Bell and Abbeduto 2007	1) DS (n=23) 2) FXS (n=18) 3) MA matched to DS & FXS (n=21)	13–24 years, M=17 12–23 years, M=17 4–7 years, M=5	Evaluative devices: density, diversity, sound effects, exclamations Evaluative devices (mental states, character name, repetition, dialogue, exaggeration)	DS > MA DS > FXS DS = MA DS = FXS
Lacroix et al. 2007	1) DS (n=12) 2) WS (n=12) 3) TD-CA (n=12) 4) MA-overall IQ (n=12)	7–19 years, M=12 7–19 years, M=12 7–19 years, M=12 4–10 years, M=7	Evaluative devices (attention-getters, emotional verbs, sound effects, dialogue)	DS = MA DS = TD-CA DS < WS
Reilly et al. 1990	1) DS matched to WS on CA, MA, sex (n=4) 2) WS (n=4) 3) MA-matched (n=4)	adolescents adolescents 7 and 8 years.	Evaluative devices (affective states, dialogue, sound effects, emphatics, attention-getters, negation[d])	DS = MA DS < WS
			THEME	
Bernicot et al. 2003	1) DS (n=7) 2) WS (n=7) 3) TD (n=7)	7–19 years, M=13 7–19 years, M=13 7–19 years, M=13	Theme maintained throughout story	DS < TD DS < WS

Table 1 (continued)

Authors	Groups (n)[a]	Age Range, Means (rounded up)	Outcome variable	Findings (comparisons involving DS groups)[b]
Hogan-Brown et al. 2013	1) DS (n=17) 2) MA (n=16) 3) FXS (n=18) 4) FXS-ASD (n=23) 5) ASD (n=20)	7–15 years, M=11 3–6 years, M=5 7–15 years, M=10 6–15 years, M=10 4–13 years, M=9	Theme maintained throughout story	DS = MA DS = FXS-AUT
Lacroix et al. 2007	1) DS (n=12) 2) WS (n=12) 3) TD-CA (n=12) 4) MA-overall IQ (n=12)	7–19 years, M=12 7–19 years, M=12 7–19 years, M=12 4–10 years, M=7	Theme included in story	DS < MA DS < TD-CA DS < WS
Miles and Chapman 2002[c]	1) DS (n=33) 2) MA (n=33) 3) MLU match (n=33) 4) SYN-COMP match (n=33)	12–26 years, M=19 2–9 years, M=5 2–9 years, M=4 2–8 years, M=5	Theme included in story CAUSALITY	DS < MA DS > MLU DS = SYN-COMP
Baron-Cohen et al. 1986	1) DS (n=15) 2) MA (n=27) 3) ASD (n=21) subset of above: 1) DS (n=7) 2) MA (n=9) 2) ASD (n=10)	6–17 years, M=11 4–6 years, M=6 6–17 years, M=12	Sequencing picture stories with: mechanical causes psychological causes Telling stories with: mechanical causes mental states (potentially but not uniquely as causes)	DS = MA; DS < ASD DS = MA; DS > ASD DS = MA; DS < ASD DS = MA; DS > ASD
Boudreau and Chapman 2000[c]	1) DS (n=31) 2) MA (n=31) 3) MLU match (n=27) 4) SYN-COMP match (n=28)	12–26 years, M=18 2–8 years, M=6 2–9 years, M=4 2–8 years, M=5	Structure of stories with causally connected events	DS = MA DS > MLU DS = SYN-COMP
Kim et al. 2008	DS (n=12) televised (TV) and aural-only conditions	6;2–7;2 years, M=6;8	Recall of stories as a function of causal connections	Recall > for stories with many connections (TV condition)

Table 1 (continued)

Authors	Groups (n)[a]	Age Range, Means (rounded up)	Outcome variable	Findings (comparisons involving DS groups)[b]
			INFERENCING	
Boudreau and Chapman 2000[c]	1) DS (n=31) 2) MA (n=31) 3) MLU match (n=27) 4) SYN-COMP match (n=28)	12–26 years, M=18 2–8 years, M=6 2–9 years, M=4 2–8 years, M=5	Inferences	DS = MA DS > MLU DS > SYN-COMP
Kay-Raining Bird et al. 2004[c]	1) DS (n=23) 2) MA (n=24)	13–20 years, M=16 4–6 years, M=5	Inferences plausible implausible	DS = MA DS > MA
Loveland et al. 1990	1) DS (n=16) 2) ASD, matched to DS on MA-verbal (n=16)	5–27 years, M=13 5–27 years, M=14	Inferences (responses to "speculative and extrapolative questions")	DS > ASD

ASD Autism Spectrum Disorder, *DS* Down syndrome, *FXS* Fragile X syndrome, *TD* Typically-developing, *CA* Chronological age, *MA* Mental age, *MLU* Mean Length of Utterance, *SYN-COMP* Syntax Comprehension

[a] All MA matching was on nonverbal tests unless otherwise specified

[b] In studies involving multiple groups, some authors did not report all between-group comparisons; this is reflected in the data reported here

[c] Studies sharing this subscript involved participants drawn from the same pool

[d] Reilly et al. also reported scores aggregating these forms of evaluation with causal connectors, inferences, and internal states, but since the data were not statistically disaggregated, the latter set of results are not reported here

problem in the story, and evaluative devices. They found no significant group differences, but MA did predict performance.

Evidence that individuals with DS have basic narrative structure is also found in Loveland et al. (1990). They reported that a DS group and a group with autism (matched on verbal age) both produced "primitive" narratives with recognizable plots. Individuals with DS have also been found to respect the order of events when memory span is controlled (Kay-Raining Bird and Chapman 1994).

However, in the studies comparing DS to MA or language matches, the matches were considerably younger (4–9 years younger), as shown in Table 1. Moreover, MA matches outperformed DS groups in some cases in terms of the number of narrative elements produced (Miles and Chapman 2002); elaboration of story schema (Lacroix et al. 2007); amount of setting information (orientation) provided (Reilly et al. 1990); and recall of story propositions (Kay-Raining Bird and Chapman 1994; Kay-Raining Bird et al. 2004).

Thus, the literature points to individuals with DS displaying basic narrative structure, but their narratives seem to lack elaboration. Their expressive narrative skills, however, continue to develop and findings by Finestack et al. (2012) point to adolescents and young adults with DS outperforming younger typically developing MA-matches in introducing characters and story settings. Furthermore, when matched on MA on an individual basis (rather than by group), they had higher total scores on a narrative measure and on subscales of conflict/resolution and cohesion. When a subset of the DS group was individually matched to the younger group on a syntactic measure, the DS group also performed as well. Additionally, the scores of the DS group and a group with FXS were not significantly different on any measure.

The findings for comprehension of story grammar were limited to one study comparing a DS group to MA matches with autism. As Table 1 shows, in Loveland et al. (1990), the DS group scored higher than the comparison group on comprehension questions about story elements and also produced fewer off-topic responses, demonstrating they had understood meaningful aspects of the story.

There is also some (albeit limited) evidence that intervention can enhance children's and adolescents' knowledge of story grammar. Schoenbrodt et al. (2009) investigated the efficacy of a parent-training program in increasing the narrative skills and vocabulary of two children with DS. Intervention components were drawn from the research literature. Parents received instruction about narrative development; materials (e.g., laminated pictures of characters); sets of questions to ask children during repeated reading sessions; reminders of temporal terms to use and elicit (e.g., first, then, finally); suggestions of art activities that entailed use of story elements; and semantic word maps to enhance vocabulary. Following intervention, the children improved on both narrative measures (story generation and story retell) and vocabulary. In a case study of an adolescent with DS, Lecas et al. (2011) capitalized on the participant's visual memory, by allowing him to use a software program to illustrate a narrative as it was told to him. The authors found that training in visually recoding spoken sentences of a short story was associated with increased information recall and comprehension. In summary, in Schoenbrodt and Lecas et al.'s studies, significant improvements were noted, suggesting that the narrative skills of individuals with DS can be supported with evidence-based practices. It should be noted, however, that these articles were the sole intervention studies we found that addressed narrative skills in DS (and the table was reserved

for studies comparing between- and within-group research). We return to this gap in the literature in the "Discussion" section of this review.

High Point and Evaluation Bernicot et al. (2003) was the only study that compared rates of evaluation by children and adolescents with DS to CA and MA matches; these groups were similar to one another, while a group with Williams Syndrome (WS) was more expressive. Hogan-Brown et al. (2013) compared the use and diversity of evaluative devices used by individuals with DS to those with FXS, FXS with autism (FXS-AUT), autism, and MA matches. Reported findings involved the DS group performance not significantly differing from those of the MA and FXS-AUT groups.

Studies of evaluation types reveal that individuals with DS use evaluative devices, but ones that are least linguistically demanding. In Reilly et al. (1990), statistical analyses found DS and MA matches to not differ in use of affective enhancers (e.g., use of sound effects). Yet performances of the DS group were significantly lower than those of individuals with WS. Use of narrative enrichment devices (i.e., affective and causal connectors) were less often used by the DS compared to the WS and MA groups. The results of Keller-Bell and Abbeduto (2007) were similar; they reported that the DS group used onomatopoeia (sound effects, e.g. "bong") as an evaluative device more than FXS groups and MA matched children. Lacroix et al. (2007) similarly found that a DS group provided comparable amounts of evaluative devices (e.g., attention getters) to MA and chronologically matched typically developing groups but less than a group with WS. Reilly et al. (1990) likewise earlier reported that similar findings with their WS and MA matches.

As the reader will recall (see Narrative Dimensions), "high points" tend to be, at least in typically developing children, marked by the use of evaluative devices; thus, one would expect low levels of evaluation to correlate with absences of high points. This prediction was confirmed by van Bysterveldt et al. (2012) who observed that individuals with DS could use evaluative devices but did not do so independently at the level recquired for marking a high point. In fact, individuals with DS produced all the components of a classic narrative according to the high point model, but needed considerable prompting and scaffolding to do so, as illustrated in Appendix 1.

Theme In the present studies, theme was construed as the main problem of the story (e.g., a boy looking for his pet frog in one of the oft-used "frog stories" in narrative research). The ability of individuals with DS to introduce the main theme or to continue referring to it was investigated in five studies: Reilly et al. (1990), Miles and Chapman (2002), Bernicot et al. (2003), Lacroix et al. (2007), and Hogan-Brown et al. (2013). Reilly et al. (1990) found that two out of four adolescents with DS failed to mention the central theme of the story. Lacroix et al. (2007) studied story theme in a larger group using a similar task to Reilly et al.. They found that while the MA- and CA-matches did not differ significantly and their scores were greater than those of a cohort with WS, the DS group performed the least well. Bernicot et al. (2003) reported similar findings regarding theme maintenance, where the DS group performed significantly lower compared to TD and WS groups. Therefore, the research shows that articulating the principal theme (here, the problem) in a story is more challenging for individuals with DS compared to MA matches and individuals with WS. These finding, however, are not unequivocal. Hogan-Brown et al. (2013), for instance, found that a DS

group was comparable in performance to MA and FXS-AUT groups with regard to theme maintenance.

Although Miles and Chapman (2002) also found that DS participants were worse than MA matches in expressing theme, the DS participants were similar to a group matched on syntax comprehension and better than a group matched on MLU. These results suggest that expression of theme depends more on cognitive levels and language comprehension than on expressive grammar.

Causal Relationships and Inferencing

Five of the studies reviewed addressed either the understanding or expression of causal relationships or recall of causally connected events. Kim et al. (2008) analyzed narrative comprehension skills in relation to presentation modality (audiovisual and audio). In the audiovisual (television) condition, children with DS recalled events with causal connections more frequently than events with fewer connections, indicating some sensitivity to causal structure. Boudreau and Chapman (2000) also found that individuals with DS were similar to both MA and language groups in comprehension of causality.

Other studies indicate, however, that children with DS do not use causal terms routinely. For example, when Baron-Cohen et al. (1986) asked children with DS to generate a story using cartoon-like drawings of a person kicking a rock and causing it to tumble downhill, most of their stories and the stories of MA matches omitted causal terms (i.e., the groups were similar but both poor). Participants in the same study were, however, moderately able to sequence pictures illustrating physical causality, again scoring similarly to typically-developing younger children. In contrast, they were less successful than MA matches at sequencing stories implying psychological causes of events (but better than children with ASD). Also, when children in van Bysterveldt et al. (2012) were prompted (with photographs) for personal narratives, only five out of 25 children expressed causal relationships between events. Thus, while children with DS show some understanding of causality, they often fail to express causal relationships in the stories they tell. Adolescents continue to appear to perform like MA matches as well. For example, Reilly et al. (1990) examined statements about causality in a larger category of "narrative enhancements" and found that adolescents with DS performed similarly to MA matches, but better than the individuals with WS.

Inferencing regarding narrative events (e.g., inferring that "the monkey was hungry" when presented with a picture of a monkey looking for bananas) (Kay-Raining Bird et al. 2004) was explored in four studies, all involving adolescents and young adults with DS. Boudreau and Chapman (2000) found that the DS group performed comparably to MA matches and better than language-matched cohorts. Kay-Raining Bird et al. (2004) likewise compared a DS group to MA matches. They found no significant group differences in the frequency of plausible inferences. However, the DS group provided relatively more implausible inferences and extraneous information. The authors suggested that DS participants might have added unnecessary information to expand their narratives and compensate for their difficulties recalling information. Loveland et al. (1990) also reported that a DS group performed higher in inferences involving speculative and extrapolative questions (e.g., providing appropriate

responses) compared to an ASD group. Finally, Reilly et al. (1990) included inferencing in their analysis of stories by adolescents with DS and WS and MA matches with mild-moderate cognitive delays. Although inferences were not analyzed separately, descriptive data suggests that the DS group performed below the other two groups. Thus, the four studies indicate that inferencing is challenging for individuals with DS, even into adolescence and early adulthood.

Microstructure

In comparison to macrostructural features, the studies reviewed reported difficulties with microstructure more consistently. Specifically, children and adolescents were reported to tell shorter stories than MA matches (Chapman et al. 1998, 2000; Lacroix et al. 2007; Reilly et al. 1990). Nonetheless, for individuals with DS, narrative length varies depending on the task (Chapman et al. 1998; Chapman 2006; Miles et al. 2006), and appears to improve with age (Cleave et al. 2012; van Bysterveldt et al. 2012).

While none of the studies focused on how well the DS group understood the vocabulary they encountered in stories, Seung and Chapman (2003) did measure recall of content words (as a general measure of story recall). They found no significant differences between DS and comparison groups matched for MA and syntax comprehension. Results were mixed on diversity of vocabulary measures (i.e., the number of different words relayed): DS groups performed lower than MA and CA matches in some cases (Bernicot et al. 2003; Boudreau and Chapman 2000), but equal to MA and language matches in others (Keller-Bell and Abbeduto 2007; Miles and Chapman 2002). In contrast, the DS groups consistently displayed more difficulty than MA matches with cohesion (Boudreau and Chapman 2000; Lorusso et al. 2007; Moore et al. 1998) as well as morphology and syntax in all reviewed studies (Bernicot et al. 2003; Boudreau and Chapman 2000; Chapman et al. 1998, 2000; Cleave et al. 2012; Hogan-Brown et al. 2013; Keller-Bell and Abbeduto 2007; Kover and Abbeduto 2010; Kover et al. 2012; Lorusso et al. 2007; van Bysterveldt et al. 2012). The sole exclusion was Fabbretti et al. (1997). However, despite these reported challenges, several studies have pointed to children with DS developing their expressive syntax over longer periods of time (Chapman et al. 1998; Thordardottir et al. 2002).

Various intervention studies have pointed to narratives potentially being suitable contexts for targeting language skills (Chapman et al. 1998) and novel word learning (Kay-Raining Bird et al. 2004); however, these benefits are not conclusive and may come at some expense. For example, individuals with DS produced less complex syntax (Kover et al. 2012) and showed more formulation difficulties (Kover and Abbeduto 2010) when producing a narrative, a finding the authors attributed to increased demands in terms of planning speech. In light of these findings, remediating syntax skills might be best addressed in other discourse contexts.

Discussion

The purpose of this review was to identify macrostructural patterns of narrative skills of children and adolescents with DS, in order to better guide interventions in clinical and school environments. Various dimensions of narrative were considered, including story

grammar, evaluation, theme, causality, inferencing, and narrative length, as well as microstructural variables including vocabulary and grammar.

Peer-reviewed studies were identified through a comprehensive search of major databases in education, psychology, health sciences, and linguistics. Most of these studies compared children with DS to younger children with typical language development (matched on mental age, as determined by tests of nonverbal ability; language; or, more rarely, chronological age) or to individuals with cognitive delays of varying genetic etiologies (often matched by mental and/or chronological age to the DS group). Studies of fictional narrative and of expression predominated; comprehension studies were rare. Longitudinal studies and studies on intervention effects were also infrequent, thus pointing to important gaps in the literature.

Macrostructure: Summary and Implications

Narrative macrostructure was found to be a relative strength for individuals with DS, resembling that of younger children matched on MA. Specifically, their performance was comparable on various story grammar measures, including introduction of characters and settings, the relay of events, episodic components and story propositions produced in retell, and expression of plot (Finestack et al. 2012; Hogan-Brown et al. 2013; Kay-Raining Bird and Chapman 1994; Kay-Raining Bird et al. 2004, 2008; Keller-Bell and Abbeduto 2007; Miles and Chapman 2002).

DS participants' expression of story theme was lower than MA-matched younger children, but similar to language matches (Miles and Chapman 2002), suggesting that language was interfering with their ability to understand and articulate the story's central problem. Finally, the research demonstrated that DS groups use evaluative devices; that is, devices that communicate the narrator's point of view about what is meaningful and salient. The rate of these was comparable to CA matches (Bernicot et al. 2003) and CA and MA-matches (Lacroix et al. 2007), but participants tended to use devices that relied least on language (e.g., sound effects and prosody).

Thus, a portion of the extant literature points to individuals with DS displaying a "skeletal narrative" that is comparable in several respects to that of younger children and thus indicative of a delayed rather than deviant pattern of development (Rice et al. 2005). This finding constitutes a good starting point for designing interventions for DS populations. Moni and Jobling (2014) have recently made similar recommendations, emphasizing the importance of narratives in school-based literacy programs for individuals with DS. Additionally, in this context, teacher training on narrative development and associated evidence-based intervention could be helpful, given that enhancing teacher knowledge has been found to impact children's outcomes (McCutchen and Berninger 1999).

Causal Relationships and Inferencing: Summary and Implications

Few of the studies investigated narrative comprehension skills in individuals with DS. In those that did, DS groups showed some basic understanding of causal relationships (Boudreau and Chapman 2000; Baron-Cohen et al. 1986; Kay-Raining Bird et al. 2004; Kim et al. 2008; Reilly et al. 1990). The topic of comprehension is important to further

explore as studies have pointed to individuals with DS having potentially greater understanding of narrative content than their expressive language abilities allow them to display (Boudreau and Chapman 2000; Chapman et al. 2000). Moreover, narrative comprehension skills might serve to facilitate narrative production in clinical settings.

Inferencing might be a particularly important area to target. First, inferencing skills were weak even in adolescents and young adults (Boudreau and Chapman 2000; Kay-Raining Bird et al. 2004; Reilly et al. 1990). Second, according to a parent survey by Trenholm and Mirenda (2006), parents of children with DS do not often ask higher-level questions, including inferential ones; while the majority reported reading books to their children, only a quarter asked their child questions about what might happen next, or why something happened. Additionally, Ricci (2011) examined the relationship between parents' book reading practices and the language of their children with DS. They found that the type of questions parents asked during book reading correlated with children's reading comprehension and receptive vocabulary.

Microstructure: Summary and Implications

Studies showed that narrative microstructure (i.e., the language used in narrative contexts), was affected in children with DS. These findings are not surprising, given that individuals with DS typically have expressive language difficulties, affecting vocabulary and even more so, syntax and morphology (Fabbretti et al. 1997; van Bysterveldt et al. 2012).

That said, these areas of persistent challenge require language interventions, and may be targeted by speech and language specialists using a variety of strategies (Buckley and Le Prèvost 2002). Narrative-based language interventions could also be used (Swanson et al. 2005). There are interventions that use stories as a context to address language challenges. As alluded to earlier in the paper, however, stories might be best for addressing comprehension and expressive vocabulary, but may be less optimal for expressive syntax.

Limitations of the Review

One of the limitations of this review was that we placed no restrictions on study designs. That is, we included studies with group, within-subject, and single-subject designs; these are designs that are generally considered to provide varying levels of evidence. However, we purposely chose to include a wide range of studies, given that research on the narratives of individuals with DS had not yet been comprehensively reviewed in the literature. The review revealed that the majority of studies nonetheless had an important strength; participants with DS were matched to other groups on nonverbal mental age, thus controlling for nonverbal cognition which could potentially confound the results. Additionally, the majority of studies included children with DS who did not have additional comorbid disabilities (e.g., autism); this, too, helps build confidence in the results and their generalizability to other individuals with DS. With respect to the intervention studies reviewed, none involved random assignment to group, and thus the conclusions one can draw are limited (i.e., other factors could have been responsible for change in the experimental group). At the same time, given that

there is so little work in this regard, the studies reviewed are an important first step, a point elaborated below.

Gaps in the Literature and Future Directions

The review demonstrated a heavy focus on fictional narratives with limited attention to personal narratives. The attention to fictional narratives may stem from the fact that such narratives can be elicited in a standard fashion using pictures and storybooks. However, the focus on fictional narratives neglects the finding that children typically tell personal narratives before fictional ones (McCabe et al. 2008). Longitudinal studies are needed to confirm that children with DS are indeed following a similar developmental trajectory; their resemblance to younger MA matches and evidence from the current review suggest that they are. In any case, the ability to tell personal stories is important for communication (for example, for telling a parent or friend about a significant event that might have occurred in their absence), and thus a good choice for intervention. Intervention could specifically involve scaffolding personal stories of events that are meaningful to children, using visual cues as prompts.

A second gap in the literature concerns intervention studies; in fact, the review revealed only two studies of this kind, involving a total of three cases. One of these was reported by Schoenbrodt et al. (2009). In that study, two parents were trained to facilitate the narratives of their children with DS, with positive results. Parental training could be an interesting avenue for future investigation. The other intervention study was conducted by Lecas et al. (2011), with an adolescent with DS. The authors found that the participants' recall and comprehension of story events was aided by visual material (video). Visual material might have capitalized upon the participant's relative strength in visual short-term memory and compensated for the deficits in verbal short-term memory that are common in DS (Chapman and Hesketh 2001). The use of photographs and pictures to elicit narratives was also observed in most of the studies examined in this review. The use of visual aids, however, to enhance narrative skills merits attention in future research.

The dearth of narrative intervention studies is consistent with Finestack's (2012) finding that few studies target the complex language abilities of children and/or adolescents with developmental disabilities. In contrast, a modest number of studies have evaluated narrative interventions for individuals with language-learning difficulties or disorders. Such studies have revealed positive outcomes, particularly for macrostructural measures (Petersen 2011), and could serve as a resource for practitioners working with DS as well.

In summary, this paper has presented the extant literature from approximately the past 30 years regarding the narrative skills of individuals with DS. Various challenges and relative strengths were reported, with overall findings pointing to individuals with DS possessing core narrative abilities that can be enhanced with appropriate intervention.

Appendix 1

Table 2 Personal narrative of a child with DS (as reported in van Bysterveldt et al. 2012, pp. 103–104)

C = child=12;6 years-old	E = examiner
< > = overlapping speech	Xx = unintelligible segments/words.
Photograph presented = dental nurse	Topic = going to the dentist
	E: <Oh look>.
C: <The dentist>.	
C: I been to a dentist before.	
	E: The dental nurse <came to school>.
C: <at the school>.	
	E: Yeah.
	E: (Tell me) tell me about that.
C: I got (I don't know) about here a tooth out [fingers in mouth while talking].	
	E: Oh yeah.
C: About here got pulled out.	
	E: You got it pulled out?
C: Yeah.	
C: Xx.	
	E: Tell me that again, slow down.
C: Xx.	
	E: You got your tooth pulled out.
C: Right up there [puts fingers in mouth].	
	E: Yeah.
C: Out.	
C: Don't know how he did it.	
C: But xx.	
	E: What do you think he did?
C: I think they got this thingy.	
C: They wriggled it.	
	E: Yeah.
C: They twisted it.	
C: And she pulled.	
C: And it was out.	
	E: Twisted and pulled and then it was out.
C: Yeah.	
	E: Mm.
C: Easy.	
	E: Well.
C: Didn't even hurt.	
C: Didn't scream or anything.	E: Wow, you are amazing.

The text is quoted verbatim from van Bysterveldt but formatted here in columns.

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Note that items marked with an asterisk are those included in the main review.

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