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Commentary

IDENTIFYING A PROTOCOL TO ASSESS LITERACY-RELATED SKILLS IN MALTESE CHILDREN AND ADOLESCENTS WITH DOWN SYNDROME

Loredana Muscat¹, Helen Grech¹, Sue Buckley²

¹Department of Communication Therapy, Faculty of Health Sciences, University of Malta, Msida, Malta

²Department of Psychology, Faculty of Science, University of Portsmouth, United Kingdom

Abstract. Acquiring the ability to read is a complex process that involves a number of skills. Recent literature confirms that many individuals with Down syndrome (DS) can acquire useful levels of reading ability which can open new opportunities. In contrast with the typically-developing (TD) population, students with DS portray an uneven pattern of development of pre-reading and reading skills. This commentary explores the aspects that need to be considered when assessing the pre-literacy and literacy abilities of students with DS in the Maltese bilingual context. Its aim is to identify the components of an assessment protocol that could be used to evaluate the literacy-related abilities of students with DS.

Keywords: Down syndrome, reading assessment, early literacy skills

Abbreviations: DS = Down syndrome; HLE = home literacy environment; LAMC = Language Assessment for Maltese Children; OME = Otitis media with effusion; PA = phonological awareness; RCPM = Raven's Coloured Progressive Matrices; SNHL = sensorineural hearing loss; TD = typically-developing; TVPS-R = Test of Visual Perception Skills-Revised; VSTM = verbal short-term memory

1 Introduction

An average of eight babies per year is born with Down syndrome (DS) in the Maltese Islands (Department of Health, Information and Research, Malta, 2015). DS is most commonly associated with intellectual disability. Individuals with DS commonly experience difficulties in acquiring new and complex information and in coping independently (Faragher & Clarke, 2014). Specific characteristics can

contribute to the difficulty in acquiring verbal language skills, thus also inhibiting, directly or indirectly, other areas such as literacy development. Characteristics include hearing acuity, cognitive skills, the home environment, phonological awareness, auditory verbal memory and visual perceptual processing. This commentary explores these aspects and identifies related assessments which may be used with individuals with DS. Clinical assessment of Maltese children and adolescents with DS is particularly challenging because of the limited number of tests standardised on the local bilingual population. Moreover, local studies exploring the performance of individuals with learning disabilities on available tests are limited. It is therefore relevant to propose an assessment protocol that may be used to investigate the literacy-related skills of Maltese children and adolescents with DS.

2 Hearing acuity

Hearing difficulties may contribute to challenges in verbal and written language development. Between 40% and 80% of children with DS are reported to have some form of hearing loss, which may be conductive, sensorineural or both (Laws & Hall, 2014; Martin et al., 2009). A conductive loss is most common in younger children with DS, primarily due to episodes of otitis media with effusion (OME) (Laws & Hall, 2014). OME is associated with speech and language delay/disorder. The fact that OME most commonly occurs at younger ages might be associated with verbal language impairment since language acquisition occurs mostly before the age of 7. Hence, children with DS might carry the repercussions of an earlier OME (Laws & Hall, 2014), particularly since there are documented long-term effects of OME, such as auditory perceptual difficulties (Bamford & Saunders, 1991). Moreover, children with DS are likely to have recurrent episodes of OME which may also extend into early adulthood. This might continue to affect speech and language development. It has been reported that the function of the middle ear can be impaired permanently in DS (Sacks & Wood, 2003). Marcell (1995) states that if OME

Correspondence to Loredana Muscat
(loredana.muscat.02@um.edu.mt)

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is untreated for a number of years, it may cause reduced mobility of the ear drum in 40% of young adults. Individuals with DS may also suffer from sensorineural hearing loss (SNHL), which is not only permanent but could actually increase with age. SNHL could be congenital or acquired. Marcell (1995) also reports that individuals with DS may have premature presbycusis, that is, early onset of acquired hearing loss as a result of age.

When assessing individuals with DS, it is of utmost importance that their hearing acuity at the time of testing is noted as this may affect performance in the skills tested. When assessment takes place in schools or homes, portable equipment to assess hearing acuity would be ideal. A hand-held paediatric audiometer used together with a sound level meter in a quiet room should allow screening of hearing abilities, although this does not replace a full diagnostic test battery.

3 Cognitive Abilities

Cognitive ability is a strong predictor of both verbal and written language development. Predominantly, individuals with DS have a mild to moderate level of cognitive impairment (e.g. Lanfranchi, Jerman & Vianello, 2009; Martin et al., 2009). Areas of cognitive functioning which are particularly underdeveloped in individuals with DS include reasoning ability, interaction with the world and theory of mind, which includes attributing mental situations, such as desiring and thinking, to oneself and to others (Abbeduto, Warren & Conners, 2007; Turner & Alborz, 2003; Javier, Cobos & Castro, 2010). These can also be influential in the process of reading acquisition, particularly in the interpretation of text. Nevertheless, memory, specifically phonological memory, is the cognitive skill that has the main impact on the development of reading (Abbeduto et al., 2007). Cognitive skills tend to vary across individuals with DS and may be influenced by other skills such as language and hearing (Frenkel & Boudrin, 2009). Moreover, cognitive characteristics are not necessarily fixed at birth and may change, especially since cognitive development is also influenced by exposure and external support. Since cognitive abilities could affect the development of literacy skills, it is crucial that they are closely monitored in individuals with DS, particularly during their early years of education.

The Raven's Coloured Progressive Matrices (RCPM) test (Raven, Court & Raven, 1990) measures non-verbal cognition. It can be used with individuals aged 5 to 11 years, although the authors indicate that the tool can be used with an older population of persons with intellectual difficulties. Studies investigating individuals with learning disabilities often use the test's raw score for the purpose of comparison to other peer groups (Van Herwegen, Farran & Annaz, 2011), which the RCPM allows. The assessment in itself is not language-dependant, so it can be easily used across different linguistic contexts. It has also been described as culturally fair on an international level (Bass, 2000). The RCPM only requires pointing to shapes and patterns, making it easier to administer if fine motor difficulties are present. Furthermore, it has been reported that the RCPM can be

used with significant confidence in comparative research involving TD participants and others with DS (Facon & Nuchadee, 2009; Fagis et al., 2011). Lanfranchi and Carretti (2012) identified 33 studies in which individuals with DS were assessed using the RCPM.

Despite the RCPM being a useful tool for evaluating the non-verbal cognition of children and adolescents with DS, it does have shortcomings. Its norms are not applicable to the local setting since no formal attempt has been made to collect normative data for the Maltese population. Moreover, the performance of Maltese individuals with learning disabilities on the RCPM has not been documented. However, since there is no other cognitive assessment that has been standardised on the local population, the RCPM may be considered in the evaluation of the cognitive abilities of Maltese individuals with DS.

4 Early Literacy Environments

Early literacy environments support the goal of reading acquisition (Al Otaiba et al., 2009). Such experiences may include a rich home literacy environment (HLE) and parent-child book reading opportunities. Although the relationship between a rich HLE and later reading success is proven, related research on children with DS is still limited. Fitzgerald, Roberts, Pierce and Schuele (1995) reported that although the homes investigated in their study were print-rich, literacy exposure was not varied and was mostly limited to shared reading. In a more widespread survey, Al Otaiba et al. (2009) found that the majority of families which included a child with DS exhibited a rich HLE, with 75% of the families stating that they owned more than 50 children's books and 60% reporting use of flashcards or other literacy material, such as magnetic letters, for 10 to 30 minutes a day. These results suggest that literacy was being given worthy importance in the homes of children with DS from the early years. It would be interesting to find out whether a similar pattern can be observed in the Maltese context since no studies on this aspect have been identified locally. It has also been indicated that although parents of children with DS do provide opportunities for literacy exposure, the type of experience was more uni-directional, with only 20-30% of participants reporting that story re-telling or expansion of stories was carried out (Trenholm & Mirenda, 2006). On the other hand, van Bysterveldt, Gillon & Foster-Cohen (2010) reported that the majority of literacy experiences focus on shared book reading, with little importance given to drawing or pre-writing activities. This suggests that parents might benefit from support and exposure to good practices which can enhance the HLE.

The Home Literacy Environment Questionnaire (Boudreau, 2005) would be relevant to exploring the HLE of individuals with DS. This questionnaire has five sections, namely Educational Background, Language Background, Reading Books, Response to Print, Interest in Letters and Additional Questions, providing an array of both quantitative and qualitative data to analyse. This questionnaire has been used by researchers in the field of literacy and DS in New Zealand (van Bysterveldt et al., 2010). Although the tool is

not standardised locally, its author has approved a Maltese version that accommodates questions specific to the local population (Boudreau, personal communication).

5 Phonological Awareness

Phonological awareness (PA) is a component of metalinguistic skills, which refers to the ability to analyse and manipulate different components of the structure of words and its sound components (Farrar, Ashwell & Maag, 2005). PA has most often been described as the primary and most important predictor of the development of reading skills (Cardoso-Martins & Frith, 2001; Pufpaff, 2009) and is applicable to different languages. Yet, its predictive strength in relation to reading ability should not be generalised in its entirety to all languages. Moreover, if a child has limited PA, this does not imply that s/he will have reading difficulties (Gillon, 2004).

The importance of PA as a predictor of success in literacy is contentious in the literature on DS. Individuals with DS have been described as being unable to develop PA (Cossu, Rossini & Marshall, 1993), being only capable of developing less competence in PA when compared to TD children (Kumar Mishra, 2007) or having limitations in specific sub-skills (Cardoso-Martin & Firth, 2001). Moreover, other studies indicate that PA does not develop as a precursor to literacy but rather as the student progresses in his/her literacy acquisition (Næss et al., 2012). Nevertheless, most studies have shown that in DS, PA development is delayed and is linked to reading acquisition (Lemons & Fuchs, 2010)

Lemons and Fuchs (2010) reviewed 20 studies which documented the development of PA in individuals with DS and compared PA skills in DS and TD populations. Evidence showed that when participants with DS were compared to reading age-matched peers, TD individuals performed better than DS participants. Yet, initial phoneme identification, rhyme judgement, letter naming and letter-sound knowledge tasks did not indicate a statistically significant difference between groups. On the other hand, a more recent study by Steele et al. (2013) found that when compared to reading age-matched peers, 26 students with DS showed significantly lower results on rhyme and phoneme matching, yet stronger letter knowledge skills. This indicates an uneven profile of phonological awareness skills, which could be dependent on both the specific skills assessed and on the assessments used. Thus, it is important to further evaluate these skills in Maltese individuals with DS, since evidence related to this area is sparse within the local context (Zahra, 2010; Wirth, 2008).

The only standardised test that could be identified to assess PA in the Maltese bilingual population is the Phonological Awareness Screen. This test forms part of the Language Assessment for Maltese Children (LAMC) battery (Grech, Franklin & Dodd, 2011). The Phonological Awareness Screen assesses five different aspects of PA: syllabification, rhyme awareness, identification of initial sounds in words, phoneme segmentation and sound-to-letter conversion. The LAMC was specifically designed to meet the language needs of children exposed to a Maltese/English bilingual environment. It allows children to be assessed in Maltese

or English, acknowledging the sociolinguistic environment that children are exposed to in Malta. However, no studies reporting use of this tool with the local DS population are available as yet.

6 Verbal Short-Term Memory

Impaired language development is associated with verbal short-term memory (VSTM) difficulties (Gathercole & Baddeley, 1990). The latter have been observed in individuals with or without DS (Abbeduto et al., 2007). Individuals with DS are reported to have a weakness in the phonological loop, which allows for the processing of verbal language (Jarrold & Baddeley, 2001; Laws, 1998). When compared to mental age-matched peers, students with DS present with an impairment in VSTM when assessed through digit and/or word span (Vicari, Marotta & Carlesimo, 2004). Different theories have been proposed to explain difficulties in VSTM. Hulme and MacKenzie (1992) imply that reduced functioning of the phonological store is at the basis of impaired VSTM in DS. Yet, Vicari et al. (2004) maintain that an impairment in the rehearsal mechanism could be the cause of poor VSTM. The rehearsal mechanism is used to rehearse the information in the phonological store to prevent it from being forgotten (Henry, 2012). Vicari, Caselli and Tonucci (2000) indicate that VSTM difficulties arise due to poor lexical-semantic capabilities. Regardless of the cause of poor VSTM, this difficulty reportedly acts as an obstacle to both language learning and language-related skills such as reading. This is especially true if a phonic-based approach to reading is used. However, it could be that poor vocabulary and language abilities are consequently affecting the successful development of VSTM. The performance of students with DS on non-word repetition, sentence imitation and word span tasks helps in better defining the relationship between VSTM and language abilities (Næss et al., 2015).

The Nonword Repetition Test (Calleja, Grech & Bamiou, 2013) could be used to evaluate non-word repetition in Maltese children and adolescents with DS. No other non-word repetition task is currently available for the Maltese population. This tool is in the process of being validated. Calleja et al. (2013) claim that the list of non-words in the test has been developed according to content validity criteria set by COST Action IS0804 (Language Impairment in a Multilingual Society: Linguistic patterns and the road to assessment) (see Chiat, 2015). Test items are language-specific, vary in syllable length, have different segmental complexity and are available in high and low word-likeness.

Another skill that taps into VSTM is sentence imitation. Children's performance on sentence imitation has been found to correlate significantly with verbal language skills, providing a picture of linguistic knowledge (Seef-Gabriel, Chiat & Dodd, 2010). Hence, a sentence imitation measure could be included in an assessment protocol for reading skills of individuals with DS. The Sentence Imitation Task (Grech et al., 2011) is the only standardised tool available for the Maltese bilingual population, with Maltese and/or English versions that can be administered. It has been standardised on Maltese children aged 2;0 to 6;0 years. The

test is also reported to predict receptive language (Grech et al., 2011) and has been used in local research studies (Calleja et al., 2013; Grech et al., 2011).

7 Visual Perceptual Processing Skills

Although reading is predominantly a visual act, research on reading difficulties with reference to visual processing is sparse. Willows, Kruk and Crocos (1993) describe research on visual processing as being dispersed both geographically and across disciplines. Relevant studies have not been reviewed adequately and theories are still debatable. Visual processing skills are generally considered to be a relative strength in individuals with DS (Klein & Mervis, 1999), particularly with respect to verbal abilities (Yang et al., 2014). This has led to focusing educational methods on these visual strengths. However, recent research is indicating contrasting results that move away from this acclaimed strength (Zahra, 2010; Yang et al., 2014). Performance on visual processing tasks in DS is varied, with visual memory being the weakest of skills. However, in the majority of studies, participants with DS performed either at par or below cognitively-matched peers (Yang et al., 2014).

The Test of Visual Perception Skills-Revised (TVPS-R) (Martin, 2006) investigates seven perceptual areas, namely visual discrimination, visual memory, spatial relationships, form constancy, sequential memory, visual figure-ground and visual closure. Although the tool is very comprehensive, it is easy and quick to administer. The tool requires students to point at their answer, through a multiple choice mode presented in large print. Therefore, it is considered ideal for students who might have difficulty with expressing their choice orally or who might have difficulty with fine motor skills. This test also allows for the assessment of the wide age range of 4;0 to 18;11 years. It can be completed in one sitting and is therefore very time-efficient. Moreover, the tool is not language constrained and can therefore be used in the local setting without the need of translation. The TVPS-R allows the comparison of results obtained with standardisation data from 2,008 students. However, normative data was collected in North America and no local data related to this test is currently available. Nevertheless, it has been used in several research studies on an international level and with the DS population (Bower & Hayes, 1994; Nandakumar & Leat, 2010; Wan et al., 2014). Therefore, the assessment performance of Maltese children and adolescents with DS could be compared to these results.

8 Conclusion

This commentary highlights a series of tests considered to be important when assessing reading-related skills in children and adolescents with DS. The proposed assessment protocol addresses the home literacy environment, phonological awareness, visual perception, sentence imitation, non-word repetition and non-verbal cognition. This protocol constitutes a feasible test battery which should depict the best possible picture of early reading skills of Maltese individuals with DS. It is recommended that this suggested

protocol is validated and also checked for its reliability. The proposed assessment battery could be used with Maltese individuals with DS in both research and clinical contexts.

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10 Conflicts of Interest

The authors report no conflicts of interest.

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